



Foetal Congenital Anomaly Diagnoses and Maternal Mental Health

Lisa Halpin

Supervised by:

Dr Julie Van Vuuren

Dr Ross White

Dr Zoe Edwards

Dr Rachel Mumford

Dr Peter Taylor

19th June 2017

Submitted in partial fulfilment of the Doctorate in Clinical Psychology, University of
Liverpool

Acknowledgements

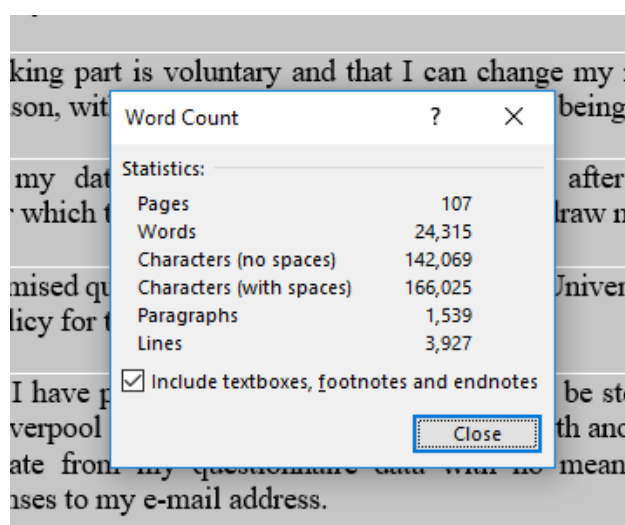
There are many people who I am indebted to for their support in the development, inception, and fulfilment of this thesis. Firstly, I would like to thank my supervisors for their continued guidance and support throughout what has been at times an uncertain journey. My thanks go to all the pregnant women who kindly participated in the study, as without them, this would not have been possible. I would like to thank the clinical nurse specialists within the regional cleft services for offering their time and advice on conducting the study. I would also like to thank my family, friends, and colleagues on the clinical psychology doctorate, for their encouragement when difficulties arose, their guidance when I felt stuck, and their kindness when I needed it the most. Finally, I would like to say a special thank you to my parents, Lorraine and Gary, for only ever asking that I try my best, and to my partner Adam, for his patience and endless support.

Table of Contents

Introductory Chapter: Thesis Overview	1
References	3
Chapter 1: Systematic Review	
Title Page	4
Abstract	5
Introduction	6
Aims	9
Method	10
Results	21
Discussion	27
Conclusion	32
References	33
Chapter 2: Empirical Paper	
Title Page	39
Abstract	40
Introduction	41
Method	46
Results	54
Discussion	61
Conclusion	70
References	71
List of Tables	
Chapter 1:	
Table 1: Systematic review search terms	11
Table 2: Quality assessment ratings	16
Table 3: Study characteristics	17
Table 4: Study aims and results	19
Chapter 2:	
Table 1: Participant characteristics	47
Table 2: Means and standard deviations for study variables	55
Table 3: Clinical cut-offs, medians, and ranges for DASS-21 score	56

Table 4: Correlation matrix: Cleft group	57
Table 5: Correlation matrix: Non-cleft group	58
Table 6: MANCOVA results	60
List of Figures	
Chapter 1:	
Figure 1: PRISMA flow diagram of articles screened and selected	13
Chapter 2:	
Figure 1: Flow of participants through the study	52
List of Appendices	
Appendix A: Author guidelines	79
Appendix B: PRISMA checklist	81
Appendix C: Systematic review protocol	83
Appendix D: Quality assessment tool	85
Appendix E: Data extraction form	87
Appendix F: Demographic information questionnaire	88
Appendix G: Measures	90
Appendix H: REC approval letter	101
Appendix I: Participant information sheet	104
Appendix J: Consent form	107

Word count: 24,315 (inc. references and appendices)



Introductory Chapter: Thesis Overview

Pregnancy is a time of physiological and psychosocial change for women, and can be a stressful life event (Hodgkinson, Smith & Wittkowski, 2014). Therefore, for some women, pregnancy can exacerbate existing psychological distress (e.g. depression, anxiety and/or stress), or contribute to its development (Biaggi, Conroy, Pawlby & Pariante, 2016). Although there are many reasons why some women experience psychological distress during pregnancy (e.g. lack of social support; Biaggi et al., 2016), this thesis is concerned with the impact of foetal congenital anomaly diagnosis on maternal mental health. Specifically, congenital heart disease (CHD) and cleft lip and/or palate (CL/P).

The original focus of the thesis was planned to be exclusively on prenatal CL/P diagnosis, however it was deemed unfeasible to conduct a systematic review in this area due to a lack of relevant quantitative research. CHD was therefore chosen as it is a commonly diagnosed congenital anomaly with sufficient literature available to conduct a systematic review. Chapter one of this thesis therefore aims to critically review, and synthesise the available literature to gain an understanding of whether prenatal CHD diagnosis is associated with maternal mental health difficulties. Clinical implications and directions for future research are considered.

The empirical paper presented in chapter two of this thesis focusses on the impact of prenatal CL/P diagnosis on maternal mental health and its associations with antenatal attachment (AA), mindfulness and self-compassion (SC). AA was selected as a variable of interest due to associations between psychological distress in pregnancy and reduced maternal-foetal attachment (Alhusen, 2008; Rubertsson, Pallant, Sydsjo, Haines & Hildingsson, 2015). Furthermore, identifying factors that might contribute to the promotion of increased AA and optimal mental health in pregnancy is therefore important. Mindfulness

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

and SC are two such factors which are increasingly demonstrating their efficacy as concepts related to reducing psychological distress and enhancing AA in pregnant women (Dunn, Hanich, Roberts & Powrie, 2012; Matvienko-Silkar, Lee, Murphy & Murphy, 2016; Mohamadirizi & Kordi, 2016). The empirical paper provides an overview of relevant research, a description of the methods used to address the research question, followed by a discussion of the results. Implications for antenatal services and directions for future research are provided.

References

- Alhusen, J. L. (2008). A literature update on maternal-fetal attachment. *Journal of Obstetric, Gynaecologic and Neonatal Nursing*, 37(3), 315-328. doi: 10.1111/j.1552-6909.2008.00241.x
- Biaggi, A., Conroy, S., Pawlby, S., & Pariante, C. M. (2016). Identifying the women at risk of antenatal anxiety and depression: A systematic review. *Journal of Affective Disorders*, 191, 62-77. doi: 10.1016/j.jad.2015.11.014
- Dunn, C., Hanich, E., Roberts, R., & Powrie, R. (2012). Mindful pregnancy and childbirth: effects of a mindfulness based intervention on women's psychological distress and well-being in the perinatal period. *Archive of Women's Mental Health*, 15, 139-143. doi: 10.1007/s00737-012-0264-4
- Hodgkinson, E. L., Smith, D. M., & Wittkowski, A. (2014). Women's experiences of their pregnancy and postpartum body image: a systematic review and meta-synthesis. *Pregnancy and Childbirth*, 14, 330. doi: 10.1186/1471-2393-14-330
- Matvienko-Silkar, K., Lee, L., Murphy, G., & Murphy, L. (2016). Effects of mindfulness interventions on prenatal well-being: A systematic review. *Psychology and Health*, 31(12), 1415-1434. doi: 10.1080/08870446.2016.1220557
- Mohamadirizi, S., & Kordi, M. (2016). The relationship between multi-dimensional self-compassion and fetal-maternal attachment in the prenatal period in referred women to Mashad Health Center. *Journal of Education and Health Promotion*, 5, 21. doi: 10.4103/2277-9531.184550
- Rubertsson, C., Pallant, J. F., Sydsjo, G., Haines, H. M., & Hildingsson, I. (2015). Maternal depressive symptoms have a negative impact on prenatal attachment – findings from a Swedish community sample. *Journal of Reproductive and Infant Psychology*, 33(2), 153-164. doi: 10.1080/02646838.2014.992009

CHAPTER 1

SYSTEMATIC LITERATURE REVIEW

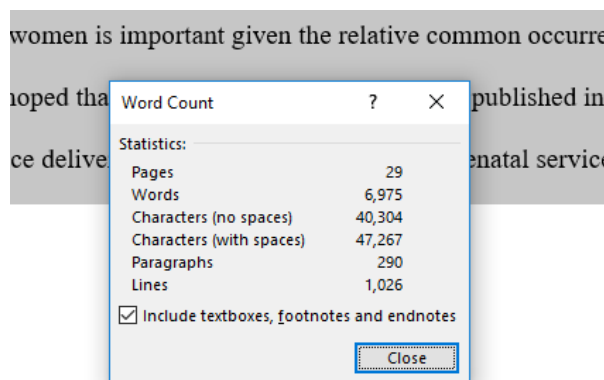
The Psychological Impact of Prenatally Diagnosed Foetal Congenital Heart Disease on Pregnant Women: A Systematic Review¹

Lisa Halpin² (MSc), Julie Van Vuuren² (BSc, DClínPsy), Ross White² (BSc, PhD, DClínPsy)
Zoe Edwards³ (MA, ClínPsyD), Rachel Mumford³ (BSc, ClínPsyD)

²University of Liverpool, UK.

³Alder Hey Children's National Health Service Foundation Trust, UK.

Word count: 6,975



Author Note

Thanks are extended to Dr Peter Taylor for his support and guidance in the early stages of this review, and to Elizabeth Bickford-Smith for offering her time as a second reviewer for the quality assessment.

¹Manuscript intended for submission to The Journal of Prenatal and Perinatal Psychology and Health (word limit 2000-8000; Appendix A)

Abstract

Congenital Heart Disease (CHD) is the most common congenital anomaly diagnosed in the foetus prenatally, yet the impact on pregnant women's mental health is not currently clear. This systematic review seeks to explore quantitative studies in this field, by examining the relationship between prenatally diagnosed CHD and aspects of maternal mental health such as anxiety, depression, and stress. PsycINFO, CINAHL, MEDLINE, SCOPUS, ETHOS and Web of Science were searched systematically using terms relating to psychological adjustment, distress, pregnancy, and foetal cardiac anomalies. Studies available in the English language and those that reported data for pregnant women were included. Seven studies met eligibility criteria and were quality assessed by two independent reviewers. Study quality was variable. Findings were qualitatively synthesised and revealed that prenatal CHD diagnosis provokes psychological distress in pregnant women. Further methodologically rigorous research is needed to allow more robust conclusions. Mandatory screening of maternal mental health following CHD diagnosis across antenatal services should be considered, in addition to developing appropriate interventions.

Keywords: congenital heart disease, pregnancy, mental health.

Introduction

Pregnancy is a time of physical, emotional, and psychological change and adjustment (Parcells, 2010). It is therefore unsurprising that some women develop mental health difficulties or experience a relapse of mental health difficulties during pregnancy (Lee et al., 2007; Raisanen et al., 2014; Ross & McLean, 2006; Smith, Shao, Howell, Lin & Yonkers, 2011). Indeed, perinatal mental health is recognised as a significant public health issue (World Health Organisation [WHO], 2008).

Specifically, the first and third trimester of pregnancy appear to be a time of increased vulnerability to anxiety and depression, which is thought to be related to the adjustment of forthcoming parental responsibilities, anxiety about the health of the child, followed later by uncertainties relating to labour and delivery (Bunevicius et al., 2009; Lee et al., 2007; Marchesi, Bertoni, & Maggini, 2009; Yanikkerem, Ay, Mutlu, & Goker, 2013).

Anxiety and depression are the most commonly experienced mental health difficulties during pregnancy (Alipour, Lamyian & Hajizadeh, 2012), and are often comorbid (Verreault et al., 2014). Although prevalence rates vary across studies, the incidence of antenatal depression has been found to range from 7-20% (see Lee et al., 2007), whilst antenatal anxiety ranges from 9-22% (see Fairbrother, Janssen, Antony, Tucker & Young, 2016). Despite these prevalence rates, pregnant women experiencing depression and/or anxiety are often over-looked by health professionals (Fairbrother, Young, Janssen, Antony & Tucker, 2015; Marcus, 2009). In addition, correctly identifying pregnant women who require support for mental health difficulties has been found to occur less than 50% of the time (Ko, Farr, Dietz & Robbins, 2012). Indeed, Goodman and Tyer-Viola (2010) found in their study, that only 15% of pregnant women identified as reaching clinical thresholds for anxiety or depression received adequate support or referral to mental health services. This is important when one considers both the short-term and long-term implications maternal mental health

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

has on the developing foetus (Lobel, Hamilton & Cannella, 2008). Research has consistently reported associations between mental health difficulties during pregnancy and outcomes such as disrupted maternal-foetal attachment, postnatal depression, premature birth, altered developmental trajectories, impaired cognitive development, and behavioural and emotional difficulties during childhood (Glover, 2011; Glover & Barlow, 2014; Huizink, Mulder, & Buitelaar, 2004; Norhayati, Hazlina, Asrenee, & Emilin, 2015; Rice, Jones, & Thapar, 2007; Ruiz & Avant, 2005; Van Den Bergh, Mennes, Mulder & Glover, 2005).

The foetal programming hypothesis (see Ellison, 2010) is increasingly used as a framework to understand how changes at critical periods in utero, because of maternal physical and mental health, can impact on the development of the foetus, and future outcomes, as described above (Cardwell, 2013). It is therefore important to understand which factors contribute to the onset and/or maintenance of psychological difficulties during pregnancy.

In their systematic review, Biaggi, Conroy, Pawlby and Pariante (2016), identified several risk factors associated with antenatal depression and/or anxiety. This included: socio-demographic and economic factors such as low income and low education level, a lack of partner and social support, a history of mental health difficulties, adverse life events, and pregnancy complications (past and present). Complicated or high-risk pregnancies can also evoke increased levels of stress and anxiety (Besser, Priel, Flett & Wiznitzer, 2007). Indeed, pregnancy-related anxiety has been well-documented in the research literature and is gaining momentum as its own specific clinical entity (Blackmore, Gustafsson, Gilchrist, Wyman & O'Connor, 2016). Furthermore, concerns relating to the foetus's health have been implicated as a central facet of pregnancy-related anxiety (Blackmore et al., 2016).

Congenital Anomalies

Springett and colleagues (2014) define congenital anomalies as: “Any defect present at delivery, probably originating before birth, and includes structural, chromosomal, genetic and biochemical defects and malformations” (p. 13). In 2012, 61% of congenital anomalies were diagnosed prenatally in the UK (Springett et al., 2014), an outcome which is often unexpected and anxiety-provoking for pregnant women (Larsson et al., 2009).

A recent study by Asplin, Wessel, Marions and Georgsson Ohman (2015), found that women carrying a baby with a prenatally diagnosed anomaly were more likely to experience depression during pregnancy and up to a year following birth than women experiencing healthy pregnancies. Furthermore, expecting a baby with an anomaly presents some women (and often their partners) with the ethical and moral dilemma of choosing to either continue with the pregnancy or have an abortion, which undoubtedly provokes more distress (Sommerseth & Sundby, 2010).

Congenital Heart Disease

The most recent report from the British and Irish Network of Congenital Anomaly Research [BINOCAR] (2014) reports that in 2012, across England and Wales, congenital heart disease (CHD) was one of the most common anomalies identified prenatally. Throughout Europe, between the years of 2011-2015, 19,889 babies were born with a CHD (European Surveillance of Congenital Anomalies [EUROCAT], n.d.). CHD severity varies and can affect different aspects of the heart structure and functioning (National Health Service [NHS], 2012). CHD severity is often categorised as mild, moderate, or severe. Whilst mild CHD and some forms of moderate CHD usually resolve within the first year of

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

life, and may not require any medical intervention, severe CHD routinely involves intrusive, and prolonged medical procedures (Solberg et al., 2012). Although medical advances have improved survival rates for CHD (Meberg, Lindberg & Thaulow, 2005), discovering that your unborn baby has a CHD is understandably stressful and upsetting for most parents (Menahem & Grimwade, 2004). Particularly, severe CHD exposes parents to a range of additional stressors such as infant survival, and stress associated with invasive medical and surgical procedures (Solberg et al., 2011; Van Horn, DeMaso, Gonzalez-Heydrich & Erikson, 2001).

Research regarding CHD diagnosis has generally focussed on the postpartum period, and highlights the ongoing experience of anxiety and depression amongst parents of infants diagnosed with CHD, even beyond conclusion of the infant's treatment (Solberg et al., 2011; Solberg et al., 2012). Despite the implications of reduced maternal mental health on the mother and developing foetus, and the knowledge that CHD diagnosis can provoke significant distress, the literature focussing solely on the psychological impact of prenatal diagnosis of CHD is significantly limited. To the author's knowledge, there have been no systematic reviews conducted to synthesise and evaluate the quality of the quantitative research conducted in this field to date.

Aims

The aim of this systematic review was to identify, synthesise, and critically evaluate the quality of existing quantitative studies exploring the relationship between prenatally diagnosed CHD and pregnant women's mental health. Conclusions will be made regarding the review findings and recommendations for future research, and clinical implications suggested.

Method

To ensure the review was undertaken systematically, the Preferred Reporting Items for Systematic Reviews and Meta-Analyses Checklist (PRISMA, 2009) was used (Appendix B). The review protocol (Appendix C) was created and registered on the international database for prospective systematic reviews in health and social care (PROSPERO: registration number: CRD42016048372).

Eligibility Criteria

This review focussed on the mental health of pregnant women who had received a prenatal diagnosis of foetal CHD. There were no restrictions regarding pregnant women's age, or publication date. Studies were excluded if they had a qualitative methodology, were not available in the English language, and if data was not available separately for pregnant women or the prenatal period. Studies were included if:

- Psychological adjustment was captured quantitatively. Use of validated measures was preferred but not essential.
- CHD was diagnosed prenatally.
- The study was available in the English language.
- Data was available separately for pregnant women, if data was also collected from other participant groups (e.g. fathers).
- Data was available independently for the prenatal period, if data was also collected at different time points (e.g. at birth).

Search Strategy

To identify relevant literature, initial scoping searches were conducted and relevant search terms were identified and combined using Boolean operators. For clarity, these were grouped into concepts and are displayed in Table 1.

Table 1.

Systematic Review Search Terms

Concept 1	Concept 2	Concept 3
Heart defect	Pregnancy	Psychological adjustment
“congenital heart disease” <i>or</i>	AND preg* <i>or</i> prenatal <i>or</i>	AND anx* <i>or</i> depress* <i>or</i>
“congenital heart defect” <i>or</i>	antenatal <i>or</i> fetal <i>or</i>	stress <i>or</i> distress <i>or</i>
“cardiac anomal*” <i>or</i>	foetal <i>or</i>	wellbeing <i>or</i>
“cardiac disease” <i>or</i>	fetus <i>or</i>	“well-being” <i>or</i>
“heart disease” <i>or</i>	foetus <i>or</i>	“life satisfaction” <i>or</i>
CHD <i>or</i>	childbearing <i>or</i>	adjustment <i>or</i>
“heart anomal*” <i>or</i>	“prenatal diagnosis”	“mental health” <i>or</i>
“heart defect”		happiness <i>or</i> “quality of life” <i>or</i> QOL

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

The electronic databases PsycINFO, CINAHL Plus, MEDLINE, Web of Science and Scopus were systematically searched for relevant published and unpublished literature from their inception until October 2016, to identify studies to include in the review. ETHOS was searched for relevant grey literature. Searches were undertaken again in May 2017, prior to completion of this review. This search revealed no additional relevant studies.

Using the specified search terms, a total of 1093 records were obtained. After the removal of 44 duplicates, the titles and abstracts of 1049 articles were screened twice by the same person. Reasons for article exclusion at this stage was predominantly related to inappropriate subject area (e.g. medical focus) and design (e.g. qualitative). Two additional studies were identified by screening the reference lists of the full-text versions of the eligible studies. Based on the inclusion and exclusion criteria, seven studies were eligible for inclusion in the final review (McKechnie, Pridham & Tluczek, 2016; Montis et al., 2007; Pinto et al., 2016; Rona, Smeeton, Beech, Barnett & Sharland, 1998; Rychik, et al., 2013; Sklansky et al., 2002; Zhu et al., 2016). Additional data, and prospective publication information and/or unpublished research were sought from the corresponding authors of included articles. Figure 1 illustrates a flow diagram of the review process.

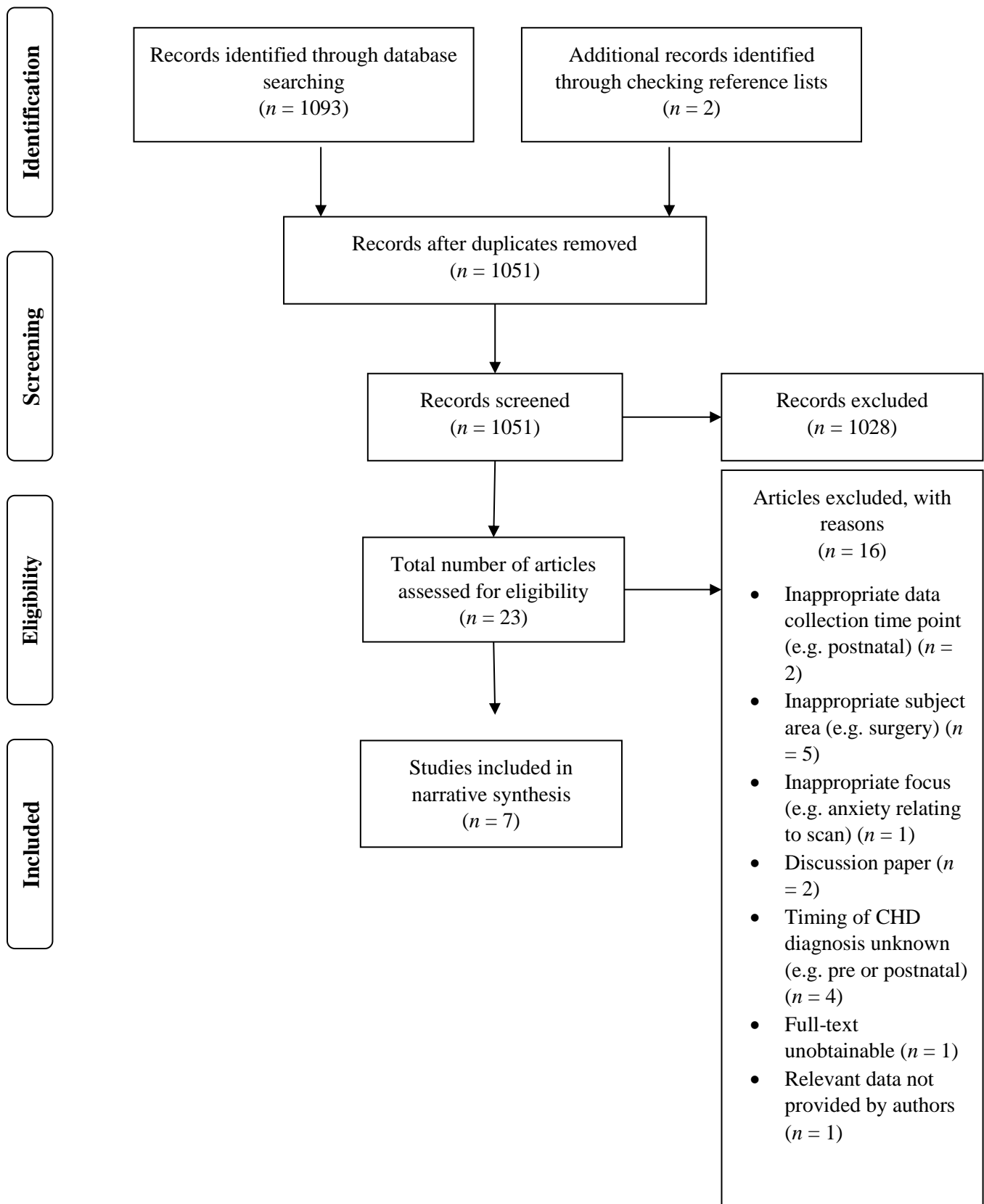


Figure 1. PRISMA diagram of study selection

Quality Assessment

Williams, Plassman, Burke, Holsinger and Benjamin's (2010) quality assessment tool of observational studies was selected and adapted to suit the context of this review (Appendix D). The tool was adapted to include eight items assessing selection bias, sample size calculation, adequate cohort description, validated methods for ascertaining psychological adjustment and CHD diagnosis, appropriate statistical analyses, acknowledgement of potential confounders/mediators, and discussion regarding missing data. Each item was rated based on the following responses: yes; no; partially; can't tell. Two reviewers independently conducted the appraisal, and any discrepancies in ratings were resolved through discussion, and liaison with a member of the research team, where applicable. Interrater reliability using Cohen's (1960) Kappa was calculated following resolved discrepancies and was .6 ($p < .001$), confirming a moderate agreement. Table 2 displays the quality assessment ratings.

The methodological quality of included studies was variable but generally poor. Two studies (Pinto et al., 2016; Rychik et al., 2013) demonstrated the most robust methodologies as they obtained the most 'yes' ratings. Three studies (Pinto et al., 2016; Rychik et al., 2013; Zhu et al., 2016) reported an unbiased selection of cohort with the remaining partially reporting unbiased selection. None of the studies provided a sample size calculation. Participant descriptions were variable across studies, although all studies provided some information. All studies used validated measures for ascertaining maternal mental health (e.g. depression, anxiety, stress), except for one study (Sklansky et al., 2002). Similarly, methods for ascertaining cardiac anomaly were adequately described in all but one study (Montis et al., 2007). One (Zhu et al., 2016) of the seven studies partially discussed missing data and the other studies did not. Three studies (Pinto et al., 2016; Rychik et al., 2013; Zhu et al., 2016) indicated that they controlled for confounding variables. All studies conducted appropriate analyses.

Data Extraction

The Centre for Reviews and Dissemination (2009) data extraction form was selected and adapted to extract relevant data from the included studies (Appendix E). Table 3 displays study characteristics and Table 4 displays study aims and results.

Study Characteristics

The predominant study design was prospective cohort (McKechnie et al., 2016; Pinto et al., 2016; Rona et al., 1998; Sklansky et al., 2002; Zhu et al., 2016), followed by cross-sectional (Montis et al., 2007; Rychik et al., 2013). Studies were conducted in the United States of America (McKechnie et al., 2016; Pinto et al., 2016; Rychik et al., 2013; Sklansky et al., 2002), United Kingdom (Rona et al., 1998), Italy (Montis et al., 2007) and China (Zhu et al., 2016). Total sample size ranged from 12 to 788. All seven studies included a comparison group(s) and reported CHD severity and/or specific diagnoses.

Mental health difficulties were measured across all studies through self-reported symptoms of depression, anxiety and/or stress/traumatic stress. In all but one study (Sklansky et al., 2002) standardised questionnaires were used. The Impact of Events Scale-Revised (IES-R; Weiss & Marmar, 1996), the Beck Depression Index-II (BDI-II; Beck, Ward, Mendelson, Mock & Erbaugh, 1961) and the State-Trait Anxiety Inventory (STAI; Spielberger, Gorsuch, Lushene, Vagg & Jacobs, 1983) were the most commonly used. Two studies (Pinto et al., 2016; Zhu et al., 2016) collected data immediately following CHD diagnosis, and five studies (McKechnie et al., 2016; Montis et al., 2007; Rona et al., 1998; Rychik et al., 2013; Sklansky et al., 2002) collected data within a specified time frame following diagnosis, ranging from 2 weeks to 10 months. Two studies did not specify the exact time point when data was collected (Montis et al., 2007; Sklansky et al., 2002).

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

Table 2

Quality Assessment Ratings

Study	Unbiased Selection of Cohort	Sample Size Calculated	Adequate Cohort Description	Validated Method for Ascertaining Psychological Adjustment	Validated Method for Ascertaining Cardiac Anomaly	Missing Data	Analysis Controls for Confounding Data	Analytic Methods Appropriate
McKechnie et al. (2016)	Partially	No	Partially	Yes	Yes	No	No	Yes
Montis et al. (2007)	Partially	No	Partially	Yes	Partially	No	No	Yes
Pinto et al. (2016)	Yes	No	Yes	Yes	Yes	No	Yes	Yes
Rona et al. (1998)	Partially	No	Yes	Yes	Yes	No	Partially	Yes
Rychik et al. (2013)	Yes	No	Yes	Yes	Yes	No	Yes	Yes
Sklansky et al. (2002)	Partially	No	Partially	Partially	Yes	No	No	Yes
Zhu et al. (2016)	Yes	No	Partially	Yes	Yes	Partially	Yes	Yes

Note. Items in boldface indicate where the reviewer's ratings were initially discrepant. Responses colour coded green = Yes; red = No; amber = Partially.

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

Table 3

Study Characteristics

Study	Design	Sample Size	Participant Characteristics	CHD Severity/Diagnosis	Measure	Assessment Times
Country						
McKechnie et al. (2016)	Prospective cohort	12	Pregnant women with a foetal CHD diagnosis ($n = 6$) and their partners ($n = 6$). All parents self-identified as married and white, 83% were college educated with household incomes between US\$30,000 and US\$90,000. The women's average age was 30 years.	Complex CHD ranging from moderate to severe.	STAI; IES-R; CES-D	4-18 weeks after diagnosis and 3-10 weeks after birth.
USA						
Montis et al. (2007)	Cross-sectional	96	Pregnant women with a foetal CHD diagnosis ($n = 48$) and their partners ($n = 48$). All couples belong to a middle social and educational status. The women's average age was 32 years. Average gestational age was 23.8 weeks.	Specific types of CHD diagnoses reported, but not categorised based on severity.	IES-R; SD	Completed during the first visit after diagnosis – exact time point not specified.
Italy						
Pinto et al. (2016)	Prospective cohort	202	Pregnant women with a foetal CHD diagnosis ($n = 60$) and their partners ($n = 59$; Group 1) and pregnant women with a postnatal CHD diagnosis ($n = 45$) and their partners ($n = 38$; Group 2). The women's average age was 28 years.	Complex CHD (defined as requiring surgery prior to newborn discharge (<30 d)).	BSI	Group 1: At time of diagnosis; Group 2: At 3 days after diagnosis; Both groups: at birth and 4-9 months follow-up.
USA						
Rona et al. (1998)	Prospective cohort	108	Pregnant women with a foetal CHD diagnosis ($n = 28$); pregnant women who received a false positive foetal CHD diagnosis ($n = 41$) and mothers of children with CHD ($n = 39$). In the foetal CHD diagnosis group, 36% of women were 25 years of age or under; 46% described themselves as a housewife/unemployed; 79% stated their religion as protestant.	Severe malformations of the heart. Specific diagnoses listed.	HADS	Between 6 and 10 months after referral to the foetal cardiology department, or after diagnosis.
UK						

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

Rychik et al. (2013)	Cross-sectional	59	Pregnant women with a foetal CHD diagnosis ($n = 59$) and pregnant women with a healthy foetus identified in the research literature. Average age 30 years; average gestational age 26 weeks; 90% were caucasian; 41% were college educated; 43% had household incomes of more than \$100,000 and 83% were married.	Serious CHD requiring neonatal evaluation and surgical or catheter based intervention within the first 6 months of life.	IES-R; BDI-II; STAI; COPE Inventory; DAS	2-4 weeks following initial diagnostic meeting.
USA						
Sklansky et al. (2002)	Prospective cohort	235	Pregnant women with abnormal or probably abnormal FE scan result ($n = 29$); pregnant women with a healthy or probably healthy FE scan result ($n = 184$) and mothers of newborns with CHD ($n = 22$). Average gestational age was 23 weeks.	CHD classified as mild (not anticipated to require cardiac catheterisation or surgery), moderately abnormal (anticipated to require cardiac catheterisation or surgery) and severely abnormal (anticipated to require surgery).	Non-standardised self-report questionnaire.	All pregnant women referred for FE, and all women with infants admitted to the neonatal intensive care unit were asked to participate. Exact data collection time points not specified.
USA						
Zhu et al. (2016)	Prospective cohort	788	Pregnant women with foetal CHD diagnosis receiving routine psychological counselling and medical self-experience counselling ^a ($n = 751$) and pregnant women with prenatal CHD diagnosis receiving routine psychological counselling ($n = 37$). Average gestational age was 24 weeks.	CHD classified as mild (no treatment required or could be easily treated), intermediate (anticipated to require surgery or cardiac catheterisation) and severe (anticipated to require cardiothoracic surgery and/or drug therapy).	STAI & BDI-II	After diagnosis; 2 weeks later; and 4 weeks later.
China						

Note. CHD = Congenital Heart Disease; STAI = State-Trait Anxiety Index; IES-R = Impact of Events Scale-Revised; SD = Semantic Differential; CES-D = Center for Epidemiologic Studies-Depression; BSI = Brief Symptom Inventory; HADS= Hospital Anxiety and Depression Scale; BDI-II = Beck Depression Index-II; DAS = Dyadic Adjustment Scale; FE = Fetal Echocardiology.

^aMedical self-experiencing counselling involved participants watching a 30-minute video of a face-to-face interview between a mother of twin girls who had undergone cardiac surgery, and a cardiac nurse, exploring the emotional experience of the mother from time of diagnosis to completion of surgery.

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

Table 4

Study Results

Study	Aims	Results
McKechnie et al. (2016)	To assess psychological well-being before and after birth following foetal CHD diagnoses.	Clinically significant levels of traumatic stress were reported for half of the pregnant women ($n = 3$) following prenatal CHD diagnosis. Compared to men, women were more likely to report clinically significant levels of distress prenatally.
Montis et al. (2007)	To analyse the emotional conditions of future mothers and fathers following CHD diagnoses.	Women had statistically significant higher total scores on the IES-R ($p < .05$) and the intrusion ($p < .05$) and arousal ($p < .05$) subscales, compared to men. CHD severity was not significantly associated with parental wellbeing scores or idealised images of participants' view of themselves as parents, or their child.
Pinto et al. (2016)	To investigate the association of timing of diagnosis with parental stress and modifiers of this relationship.	In the prenatally diagnosed group, women scored significantly higher in all domains than men: anxiety ($p < .001$); depression ($p < .01$); global psychological stress score ($p < .001$). Higher gestational age at diagnosis was associated with higher anxiety and global stress scores. Women in the prenatally diagnosed group had lower adjusted mean anxiety scores and trended towards lower global stress scores at time of birth than women postnatally diagnosed.
Rona et al. (1998)	To assess levels of anxiety and depression in three groups of women (pregnant women with CHD diagnosis; pregnant women with false-positive diagnosis and women with an affected child).	Confirmed prenatal diagnosis of foetal CHD was found to be a significant factor affecting anxiety when all three groups were analysed ($p < .01$). Women in the prenatally confirmed CHD group, and women with a child with CHD were significantly more anxious than those in the false-positive group (all $p < .01$).

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

Rychik et al. (2013)	To determine whether prenatal diagnosis of CHD increases maternal stress.	When compared to pregnant women of a similar age, with a healthy foetus, participants in this study experienced significantly higher depression ($p < .003$) and anxiety ($p < .0001$) (state and trait scores). Scores exceeding clinical thresholds were found in women expecting a baby with CHD for depression (22%), trait anxiety (14%), state anxiety (31%) and traumatic stress (39%).
Sklansky et al. (2002)	To examine the psychological impact of both normal and abnormal FE.	This study revealed that 21% of women with an abnormal FE result felt happier as a result of the FE, and 24% felt less happy as a result of FE, compared to 61% of women in the healthy pregnancy group who felt happier as a result of FE ($p < .015$). Forty-eight percent of women with abnormal FE felt more anxious as a result of FE compared to 5% of women in the healthy pregnancy group ($p < .001$).
Zhu et al. (2016)	To assess maternal psychological responses after echocardiographic detection of a suspected fetal heart anomaly.	Women in the severe CHD group had significantly higher state anxiety, trait anxiety, and depression scores than women in the mild and intermediate CHD groups (all $p < .05$). Compared to healthy controls (data from a prior study), women in all three CHD categories had significantly higher anxiety and depression scores (all $p < .01$). When scores exceeding clinical thresholds were analysed, the largest percentages were found in the severe CHD group (87% state anxiety; 84% trait anxiety and 91% depression). State anxiety and depression scores were significantly higher in the severe CHD group (all $p < .05$).

Study Results

All studies explored the impact of prenatal CHD diagnosis on the mental health of pregnant women. Some studies also explored additional factors in relation to mental health which included: CHD severity, timing of CHD diagnosis, change over time, socioeconomic variables, coping skills, and subsequent perception of self as a parent and perceived child characteristics. All studies reported statistical and/or clinically significant levels of psychological distress in pregnant women with a prenatal CHD diagnosis, however, sample size, data collection time-points, comparison groups, and CHD severity/categorisation varied across all studies. Although each study offers a unique insight into the experiences of pregnant women with a CHD diagnosis, variability in quality assessment ratings limits the strength of conclusions that can be drawn from the findings. Interestingly, only one study (Pinto et al., 2016) and one of the most methodologically robust studies, found that women prenatally diagnosed had lower anxiety and global stress scores when compared to women postnatally diagnosed.

In studies that compared pregnant women and their partners, women generally reported increased levels of psychological distress. McKechnie et al. (2016) found that more women than men met clinical thresholds for traumatic stress, anxiety, and depression following CHD diagnosis. However, this study was based on a *very* small sample of white, married couples from high earning households which limits inferences that can be drawn and overall generalisability of findings. The timing at which data was collected was also variable between participants which further limits the strength of conclusion drawn. For example, participants who had completed measures 4 weeks after diagnosis would perhaps be expected to experience greater levels of distress compared to participants completing measures at 18 weeks' post-diagnosis. Additionally, CHD diagnosis was classified as moderate to severe which perhaps contributed further to elevated scores. Similarly, Montis et al. (2007) found

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

that women had significantly higher levels of overall traumatic stress compared to men and significantly higher scores on intrusion (difficulties averting attention from the diagnosis) and arousal (feelings of frequent tension). However, in this study, the point at which data was collected from participants was not specified which undoubtedly affects the reliability of conclusions that can be drawn. Furthermore, the average gestational age was 23.8 weeks which is perhaps an indication that participants had only recently received a CHD diagnosis which again might skew findings if participants are still adjusting to the diagnosis. Pinto et al. (2016) also found that women's scores were significantly higher than men's following prenatal CHD diagnosis, on depression, anxiety and global stress scores. However, although this study recruited a greater number of participants than McKechnie et al. (2016) and Montis et al. (2007) they only recruited participants who had received a *complex* CHD diagnosis which would involve surgery within the first 30 days following birth. Undoubtedly, without a control group of women experiencing different severities of CHD diagnosis, this limits the conclusions made by Pinto et al. (2016).

When compared to pregnant women expecting a healthy baby, pregnant women with a foetal CHD diagnosis reported feeling significantly more anxious, and less happy following foetal echocardiology (Sklansky et al., 2002). Findings from the Sklansky et al. (2002) study however, are based on data from a non-standardised measure which was designed by the study author. It is therefore difficult to make any reliable conclusions from this study.

Furthermore, Rychik et al. (2013) found that women expecting a baby with a CHD reported significantly higher depression, anxiety (state and trait) and traumatic stress scores, than women pregnant with a healthy baby. Again, the CHD diagnosis was classified as severe requiring surgical intervention within the first 6 months of life and it is therefore unsurprising that differences were found between these two groups of women.

CHD severity.

When severity of CHD was explored, Montis et al. (2007) did not find that it was significantly associated with psychological wellbeing in pregnant women. However, they did not report CHD severity based on categories (i.e. mild; moderate; severe) and therefore might have overlooked subtle differences between participants. In contrast, Zhu et al. (2016) found that when compared to women with a mild or intermediate foetal CHD diagnosis, women with a foetal diagnosis of severe CHD exhibited significantly higher state anxiety and depression scores. Furthermore, women in the severe CHD diagnosis group also reported the highest percentage of clinically significant depression and anxiety (state and trait) scores. Due to the large sample size, use of standardised measures and more stringent data collection time points and explicit CHD categories, the results of this study offer more robust conclusions. However, this study is limited by the fact that it did not employ a control group of women who did not have a CHD diagnosis and did not discuss in detail participant demographics.

Timing of diagnosis.

When timing of diagnosis was explored (e.g. prenatal vs. postnatal) Pinto et al. (2016) found that women who received a prenatal CHD diagnosis were not significantly different on any domain score (i.e. global stress, anxiety, depression) compared with women diagnosed postnatally. Interestingly, women who received a prenatal diagnosis had lower anxiety and global stress scores compared to women diagnosed postnatally, when assessed at birth. Perhaps a prenatal diagnosis gave women time to adjust well in advance of their baby's birth.

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

In contrast, Rona et al. (1998) found that women with a true positive (confirmed CHD) prenatal diagnosis had higher levels of anxiety than women with a false positive (suspected CHD but not confirmed by a specialist) diagnosis. Women who gave birth to a child with CHD who were not expecting this diagnosis (e.g. false negative diagnosis) expressed higher levels of clinically significant anxiety and depression, than women who received either a true positive or false positive CHD diagnosis. However, in this study, participants were recruited from the same hospital site indicating a selection bias and demographic information was only collected for women prenatally diagnosed. This further limits the conclusions that can be made because potential confounding variables, such as socioeconomic status have not been accounted for in the analysis.

Change over time.

When the psychological impact of prenatal CHD diagnosis was tracked over time, McKechnie et al. (2016) found that half ($n = 3$) of pregnant women in their sample reported clinically significant levels of traumatic stress at time of diagnosis, which only remained the same for one pregnant woman at time of birth. Clinical cut-offs were also met for depression and anxiety, for two of the three women prenatally, and following birth, both women reported clinically significant depression scores. However, data was collected 3-10 weeks after birth which limits comparison between the participants. Conversely, Pinto et al. (2016) found that women who had received a prenatal diagnosis maintained significantly lower levels of anxiety and global stress scores at birth, than women postnatally diagnosed. At follow-up, no differences existed between the two groups. However, follow-up data was captured at some point between four and nine months after birth, which could have affected findings between the two groups. For example, if participants who had been diagnosed at birth only completed

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

the outcome measure nine months later, the result might have been different if they had completed four months after giving birth and receiving the diagnosis.

Zhu et al. (2016) also explored change over time; however, this was related to the effect of a counselling intervention. Zhu et al. (2016) found that women in the mild and intermediate CHD diagnosis group had a statistically significant reduction in anxiety and depression scores following medical self-experiencing counselling. No significant differences were found for women in the severe CHD diagnosis group following any intervention. However, as the highest percentages of clinically significant scores were found in the severe CHD diagnosis group, perhaps this is an indication that participants in this group were unable to engage in an intervention relatively soon after diagnosis.

Socioeconomic variables.

Rychik et al. (2013) found that pregnant women with a prenatal CHD diagnosis reported significantly lower partner satisfaction than women experiencing a healthy pregnancy. Lower partner satisfaction and lower income was found to be significantly associated with higher depression scores. However, this study was based on a relatively small sample size with unknown statistical power whereby participants were compared to a 'control group' identified in the literature. Failure to employ a 'live' control group of women for the study seriously limits any conclusions drawn and further generalisability. Rona et al. (1998) found that younger women, those who paid more for transport to the hospital site and those whose religion was other than protestant had significantly higher levels of anxiety. Higher gestational age at diagnosis was also associated with higher anxiety and stress in women with prenatally diagnosed CHD (Pinto et al., 2016).

Coping skills.

Rychik et al. (2013) examined the role of coping skills and found that statistically significant relationships existed between depression, state anxiety, traumatic stress and the coping variables acceptance, denial, positive reinterpretation and the use of social support. Specifically, higher acceptance was associated with lower depression and lower anxiety, and higher scores in positive reinterpretation and growth were significantly associated with lower anxiety. Higher denial was associated with higher depression, higher anxiety and higher traumatic stress. However, based on the limitations of this study, as stated in the preceding paragraph, conclusions made relating to these identified associations are limited.

Other factors.

Montis et al. (2007) reported that pregnant women expecting a baby with a CHD felt significantly more insecure and anxious in their image of themselves as parents compared to men, and imagined their baby to be shyer, more fearful, sadder and less independent. However, this study only partially provided an adequate description of the cohort and information pertaining to previous mental health difficulties and social support was not reported. This could potentially impact on pregnant women's image of themselves as parents and subsequently of their child.

Discussion

The aim of this review was to identify quantitative studies addressing the association between prenatal CHD diagnosis and maternal mental health. All of the included studies reported statistically and/or clinically significant associations between prenatal CHD diagnosis and the self-reported experience of depression, anxiety, stress, and/or traumatic stress in pregnant women. The findings of this review are consistent with previously published research highlighting associations between prenatally diagnosed CHD and psychological distress (Menahem & Grimwade, 2004; Solberg et al., 2012). These findings also support the association between prenatal diagnoses of congenital malformations generally, and experiencing increased levels of psychological distress (Asplin et al., 2015; Larsson et al., 2009).

When studies examined specific variables thought to contribute to psychological distress, findings were variable. Regarding severity of CHD, one study in this review did not find that it was significantly associated with psychological distress in pregnant women. However, partial support was found for Van Horn et al. (2001) who highlighted the impact of CHD severity on increased psychological distress. Similarly, partial support was found for Biaggi et al. (2016) and Besser et al. (2007), in relation to the impact of timing of diagnosis on maternal psychological wellbeing. Interestingly Pinto et al. (2016) found that women prenatally diagnosed were in fact less stressed and less anxious than women postnatally diagnosed. Pinto et al. (2016) felt that this finding might reflect the positive influence of additional support from health professionals that women in their study received.

This review also highlighted other variables impacting on women's mental health. This included partner satisfaction, income, maternal age, religion, gestational age at diagnosis and coping skills (e.g. acceptance, denial, positive reinterpretation and growth).

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

Identification of variables such as this indicates the multifaceted nature of factors affecting psychological wellbeing in pregnant women.

Strengths and Limitations of Included Studies

The quality assessment highlighted the methodological variability between studies and the limitations within each study. Although some interesting findings have been reported, caution must be used when considering the strength of the conclusions made. The main issues are highlighted and discussed below.

Most of the reviewed studies used well-validated, self-report measures. However, because of the variety of measures used across studies, and the bias inherent in self-report methods, the overall confidence in comparability across studies and the conclusions that can be drawn is limited. However, given the nature of this research, self-report measures are perhaps the most practical and economical method to collect data as opposed to conducting clinical interviews. Cross-sectional research also limits inferences that can be drawn from findings.

Only two studies recruited a control group of pregnant women expecting a healthy foetus, and one of these control groups was in fact secondary data that had been collected in a pre-existing study. Lack of appropriate control groups further limits interpretations that can be made from the reported findings, as it cannot be confidently concluded that the observed effect was a result of the CHD diagnosis. Furthermore, four of the included studies recruited a sample of pregnant women with severe or complex CHD diagnoses, again limiting generalisability of findings.

General measures of anxiety and depression were used in the included studies rather than pregnancy-specific measures, such as the Pregnancy Related Anxiety Questionnaire –

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

Revised 2 (PRAQ-R2; Huizink, et al., 2016). In their recent systematic review, Brunton, Dryer, Saliba and Kohlhoff (2015) highlight the questionable use of measures such as the Hospital Anxiety and Depression Scale (HADS; Zigmond & Snaith, 1983) and STAI (Spielberger et al., 1983) in pregnant samples, which have been validated based on different samples and constructs. This perhaps further limits the strength of conclusions that can be drawn. However, most of the included studies collected data postnatally and involved a range of participants (e.g. not just pregnant women), which allowed for more meaningful group comparisons to take place.

Not all studies reported gestational age or discussed its influence as an important factor relating to psychological vulnerability and pregnancy-related anxiety as previously demonstrated (e.g. Bunevicius et al., 2009; Lee et al., 2007; Marchesi et al., 2009; Yanikkerem et al., 2013). Additionally, none of the included studies controlled for mental health difficulties that predated the CHD diagnosis, or accounted for a history of mental health difficulties. This is an important factor to consider as research has previously demonstrated (Lee et al., 2007; Raisanon et al., 2014).

Sample size was variable across studies, which has implications relating to statistical power and the avoidance of reporting a false positive or false negative result. Related to this, none of the studies provided power calculations which leaves doubt regarding whether adequate power was achieved. Reporting of participant demographics was also limited; however, some studies did acknowledge limited ethnic diversity amongst participants, which affects generalisability, and overall conclusions made in this review. Included studies generally studied pregnant women who had received a diagnosis of complex/severe CHD; this further skews the findings and limits interpretation.

Strengths and Limitations of the Current Review

The use of comprehensive search strategies allows for a good degree of confidence that all relevant research was identified and included. The current review was based on a comprehensive and reproducible protocol, stipulating clear inclusion and exclusion criteria (Appendix C). The quality of included studies was assessed independently by two reviewers. In hindsight, it would have been advantageous to have also involved a second reviewer in the initial screening and selection process. Limiting this review to quantitative research only, and to studies only available in English, further reduces the amount of relevant research captured, therefore introducing language and cultural biases.

Two studies that met eligibility criteria could not be included in the review. Despite numerous attempts, the full-text version by Davey et al. (2016) study was not obtained within the time frame of this review. Additionally, a study by Brosig, Whitstone, Frommelt, Frisbee, and Leuthner (2007), was also excluded from this review as the required data (i.e. separate data for women) was not provided by the corresponding author. The failure to include these studies may serve to diminish the extent to which the current review can be considered a comprehensive consideration of all the relevant research.

Implications for Future Research

It is clear from the limited number of studies in this review, that this topic area warrants further investigation. Reasons for why this area is comparatively under-researched, are unknown to the author, however it is felt that this is a valuable topic to be explored, given the impact of maternal wellbeing on foetal development and outcomes.

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

Future research would benefit from including additional comparison groups (e.g. other congenital anomalies) and routinely recruiting a control group of women experiencing healthy pregnancies. It would also be helpful to explore factors associated with resilience, and coping styles. Studies exploring the efficacy of psychological interventions during pregnancy following prenatal CHD diagnosis would also be advantageous, particularly those employing a longitudinal and randomised design.

Quantitative research would benefit from employing consistent and validated measures assessing mental health. Ensuring appropriate statistical power is met by conducting power calculations and recruiting larger samples would also be advantageous. Identifying potential confounding and/or mediating variables, and accounting for these in the study design and statistical analyses would also improve the strength of conclusions that can be drawn. Recruiting demographically diverse and ethnically diverse samples would also be beneficial in future research, as it has been highlighted previously that ethnic minorities often experience increased levels of stress during pregnancy (Robinson, Cairns, Cairns, Fung & Tough, 2016).

Implications for Professional Practice

This review highlights that a prenatal CHD diagnosis might provoke and/or contribute to psychological distress in pregnant women. Offering formal (e.g. clinical interview) and informal (e.g. routine psychometrics) psychological assessment and/or screening to pregnant women following a prenatal CHD diagnosis would therefore be beneficial. Furthermore, identifying pregnant women who are at increased risk of experiencing psychological distress following a prenatal CHD diagnosis, due to certain socioeconomic variables (e.g. low income), would be advantageous.

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

All health professionals working in antenatal services can offer basic psychological screening, psycho-education and sign-posting to relevant mental health services and charities. Providing evidence-based interventions to individuals and groups, focussing on promoting coping skills and enhancing resilience would be beneficial. Delivering consultation and training to staff would also help promote awareness of mental health difficulties associated with prenatal CHD diagnosis, and improve the identification of women in need of support. Furthermore, peer support opportunities from people who have been through this experience would also be helpful.

Conclusion

The results of this review highlight that the relationship between prenatal CHD diagnosis and the psychological wellbeing of pregnant women is a complex one, which requires further exploration. Although included studies illustrated an association between prenatal CHD diagnosis and symptoms of depression, anxiety, and traumatic stress, the methodological limitations of included studies reduce confidence in the conclusions drawn. It is important for additional research to be conducted within this population and to further examine the role of prenatal CHD diagnosis and the numerous medical (e.g. severity of CHD), psychosocial (e.g. social support) and pregnancy-specific variables (e.g. wellbeing factors associated solely with pregnancy) that may contribute to reduced maternal mental health.

Understanding the impact of prenatal CHD diagnosis and associated psychological sequelae for pregnant women is important given the relative common occurrence of prenatal CHD diagnosis. It is hoped that more research is conducted and published in this area, to support effective service delivery and policy development in antenatal services, where appropriate.

References

- Alipour, Z., Lamyian, M., & Hajizadeh, E. (2012). Anxiety and fear of childbirth as predictors of postnatal depression in nulliparous women. *Women and Birth: Journal of the Australian College of Midwives*, 25(3), e37-e43. doi: 10.1016/j.wombi.2011.09.002
- Asplin, N., Wessel, H., Marions, L., & Georgsson Ohman, S. (2015). Maternal emotional wellbeing over time and attachment to the fetus when a malformation is detected. *Sexual and Reproductive Healthcare*, 6(3), 191-195. doi: 10.1016/j.srhc.2015.04.003
- Beck, A. T., Ward, C. H., Mendelson, M., Mock, J., & Erbaugh, J. (1961). An inventory for measuring depression. *Archives of General Psychiatry*, 4, 561-571.
- Besser, A., Priel, B., Flett, G. L., & Wiznitzer, A. (2007). Linear and nonlinear models of vulnerability to depression: personality and postpartum depression in a high risk population. *Individual Differences Research*, 5(1), 1-29.
- Biaggi, A., Conroy, S., Pawlby, S., & Pariante, C. M. (2016). Identifying the women at risk of antenatal anxiety and depression: A systematic review. *Journal of Affective Disorders*, 191, 62-77. doi: 10.1016/j.jad.2015.11.014
- Blackmore, E., Gustafsson, H., Gilchrist, M., Wyman, C., & G O'Connor, T. (2016). Pregnancy-related anxiety: Evidence of distinct clinical significance from a prospective longitudinal study. *Journal of Affective Disorders*, 197, 251-258. doi: 10.1016/j.jad.2016.03.008
- British and Irish Network of Congenital Anomaly Research. (2014). *Congenital anomaly statistics 2012 England and Wales*. Retrieved from http://www.binocar.org/content/Annual%20report%202012_FINAL_nologo.pdf
- Brosig, C.L., Frommelt, M. A., Leuthner, S. R., Whitstone, B. N., & Frisbee, S. J. (2007). Psychological distress in parents of children with severe congenital heart disease: the impact of prenatal versus postnatal diagnosis. *Journal of Perinatology*, 27(11), 687-692. doi: 10.1038/sj.p.7211807
- Brunton, R. J., Dryer, R., Saliba, A., & Kohlhoff, J. (2015). Pregnancy anxiety: A systematic review of current scales. *Journal of Affective Disorders*, 176, 24-34. doi: 10.1016/j.jad.2015.01.039
- Bunevicius, R., Kusminskas, L., Bunevicius, A., Nadisauskiene, R. J., Jureniene, K., & Pop, V. J. M. (2009). Psychosocial risk factors for depression during pregnancy. *Acta Obstetricia Gynecologica Scandinavica*, 88(5), 599-605. doi: 10.1080/00016340902846049
- Cardwell, M. S. (2013). Stress: pregnancy and considerations. *Obstetrical and Gynaecological Survey*, 68(2), 119-129. doi:10.1097/OGX.0b013e31827f2481
- Centre for Reviews and Dissemination. (2009). *Systematic Reviews: CRD's guidance for undertaking reviews in health care*. Retrieved from University of York website: https://www.york.ac.uk/crd/SysRev/!SSL!/WebHelp/SysRev3.htm#1_3_UNDERTAKING_THE_REVIEW.htm

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

- Cohen, J. (1960). A coefficient of agreement for nominal scales. *Educational and Psychological Measurements*, 20(1), 37-46. doi: 10.1177/001316446002000104
- Davey, B., Tian, Z., Ejembi, A., Donaghue, D., Combs, J., Elci, O., Levy, S., Rychik, J., & Diamond, G. (2016). Psychological stress and maternal cortisol when carrying a foetus with congenital heart disease. *Journal of the American College of Cardiology*, 67(13), 921. doi: 10.1016/S0735-1097(16)30922-6
- Ellison, P. T. (2010). Fetal programming and fetal psychology. *Infant and Child Development*, 19(1), 6-20. doi: 10.1002/icd.649
- European Surveillance of Congenital Anomalies [EUROCAT] (n.d.). *Congenital anomaly prevalence rates 2011-2015*. Retrieved from <http://www.eurocat-network.eu/accessprevalencedata/prevalencetables>
- Fairbrother, N., Janssen, P., Antony, M. M., Tucker, E., & Young, A. H. (2016). Perinatal anxiety disorder prevalence and incidence. *Journal of Affective Disorders*, 200, 148-155. doi: 10.1016/j.jad.2015.12.082
- Fairbrother, N., Young, A. H., Janssen, P., Antony, M. M., & Tucker, E. (2015). Depression and anxiety during the perinatal period. *BioMedCentral Psychiatry*, 15(1), 206-214. doi: 10.1186/s12888-015-0526-6
- Glover, V. (2011). Annual research review: Prenatal stress and the origins of psychopathology: An evolutionary perspective. *Journal of Child Psychology and Psychiatry and Allied Disciplines*, 52(4), 356-367. doi: 10.1111/j.1469-7610.2011.02371.x
- Glover, V., & Barlow, J. (2014). Psychological adversity in pregnancy: what works to improve outcomes? *Journal of Children's Services*, 9(2), 96-108. doi: 10.1108/jcs-01-2014-0003
- Goodman, J. H., & Tyer-Viola, L. (2010). Detection, treatment, and referral or perinatal depression and anxiety by obstetrical providers. *Journal of Women's Health*, 19(3), 447-490. doi: 10.1089/jwh.2008.1352
- Huizink, A. C., Delforterie, M. J., Scheinin, N. M., Tolvanen, M., Karlsson, L., & Karlsson, H. (2016). Adaption of pregnancy anxiety questionnaire- revised for all pregnant women regardless of parity: PRAQ-R2. *Archive of Women's Mental Health*, 19, 125-132. doi: 10.1007/s00737-015-0531-2
- Huizink, A. C., Mulder, E. J. H., & Buitelaar, J. K. (2004). Prenatal stress and risk for psychopathology: Specific effects or induction of general susceptibility? *Psychological Bulletin*, 130(1), 115-142. doi: 10.1037/0033-2902.130.1.115
- Ko, J.Y., Farr, S. L., Dietz, P. M., & Robbins, C. L. (2012). Depression and treatment among U.S. pregnant and non-pregnant women of reproductive age, 2005–2009. *Journal of Women's Health*, 21(8), 830–836. doi: 10.1089/jwh.2011.3466
- Larsson, A. K., Svalenius, E. C., Ekelin, M., Nyberg, P., Dykes, A., & Marsal, K. (2009). Parent's worried state of mind when fetal ultrasound shows an unexpected finding: A

- comparative study. *Journal of Ultrasound in Medicine*, 28(12), 1663-1670. doi: 10.7863/jum.2009.28.12.1663
- Lee, A. M., Lam, S. K., Sze Mun Lau, S. M., Chong, C. S. Y., Chui, H. W., & Fong, D. Y. T. (2007). Prevalence, course, and risk factors for antenatal anxiety and depression. *Obstetrics and Gynaecology*, 110(5), 1102-1112. doi: 10.1097/01.AOG.0000287065.59491.70
- Lobel, M., Hamilton, J. G., & Canella, D. T. (2008). Psychosocial perspectives on pregnancy: Prenatal maternal stress and coping. *Social and Personality Psychology Compass*, 2(4), 1600-1623. doi: 10.1111/j.1751-9004.2008.00119.x
- Marchesi C., Bertoni, S., & Maggini, C. (2009). Major and minor depression in pregnancy. *Obstetrics and Gynaecology*, 113(6), 1292–1298. doi: 10.1097/AOG.0b013e3181a45e90
- Marcus, S. M. (2009). Depression during pregnancy: rates, risks and consequences – Motherisk Update 2008. *Canadian Journal of Clinical Pharmacology*, 16(1), e15-e22.
- McKechnie, A. C., Pridham, K., & Tluczek, A. (2016). Walking the “emotional tightrope” from pregnancy to parenthood: Understanding parental motivation to manage health care and distress after a fetal diagnosis of complex congenital heart disease. *Journal of Family Nursing*, 22(1), 74-107. doi: 10.1177/1074840715616603
- Meberg, A., Lindberg, G., & Thaulow, E. (2005). Congenital heart defects: The patients who die. *Acta Paediatrica, International Journal of Paediatrics*, 94(8), 1060-1065. doi: 10.1080/08035250410025618
- Menahem, S., & Grimwade, J. (2004). Counselling strategies in the prenatal diagnosis of major heart abnormality. *Heart Lung and Circulation*, 13(3), 261-265. doi: 10.1016/j.hic.2004.06.009
- Montis, S., Secchi, G., Mura, S., Atzeni, C., Portas, E., Congiu, M., Pisanu, S., & Tumbarello, R. (2007). Psychological issues in the diagnosis of fetal cardiac disease. *Minerva Psichiatrica*, 48, 373-378.
- National Health Service (2012). *Congenital Heart Disease: information for parents*. Retrieved from https://www.gov.uk/government/uploads/system/uploads/attachment_data/file/399199/Congenital_heart_disease_-_Parents_-_FASP76.pdf
- Norhayati, M. N., Hazlina, N. H., Asrenee, A. R., & Emilin, W. M. A. (2015). Magnitude and risk factors for postpartum symptoms: A literature review. *Journal of Affective Disorders*, 175, 34–52. doi: 10.1016/j.jad.2014.12.041
- Parcells, D. A. (2010). Women’s mental health nursing: depression, anxiety and stress during pregnancy. *Journal of Psychiatric and Mental Health Nursing*, 17(9), 813-820. doi: 10.1111/j.1365-2850.2010.01588.x
- Pinto, N. M., Weng, C., Sheng, X., Simon, K., Byrne, J. B., Miller, T., & Puchalski, M. D. (2016). Modifiers of stress related to timing of diagnosis in parents of children with

- complex congenital heart disease. *Maternal-Fetal and Neonatal Medicine*, 29(20), 3340-3346. doi: 10.3109/14767058.2015.1125465
- Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA). (2009). *PRISMA 2009 Checklist*. Retrieved from <http://www.prisma-statement.org/>
- Raisanen, S., Lehto, S. M., Nielsen, H. S., Gissler, M., Kramer, M. R., & Heinonen, S. (2014). Risk factors for and perinatal outcomes of major depression during pregnancy: A population based analysis during 2002-2010 in Finland. *British Medical Journal Open*, 4(11). doi: 10.1136/bmjopen-2014-004883
- Rice, F., Jones, I., & Thapar, A. (2007). The impact of gestational stress and prenatal growth on emotional problems in offspring: A review. *Acta Psychiatrica Scandinavica*, 115(3), 171-183. doi: 10.1111/j.1600-0447.2006.00895.x
- Robinson, A. M., Cairns, S. L., Benzie, K. M., Fung, T., & Tough, S. C. (2016). Who is distressed? A comparison of psychosocial stress in pregnancy across seven ethnicities. *BMC Pregnancy and Childbirth*, 16(1), 215. doi: 10.1186/512884-016-1015-8
- Rona, R. J., Smeeton, N. C., Beech, R., Barnett, A., & Sharland, G. (1998). Anxiety and depression in mothers related to severe malformation of the heart of the child and foetus. *Acta Paediatrica*, 87(2), 201-205. doi: 10.1111/j.1651-2227.1998.tb00976.x
- Ross, L. E., & McClean, L. M. (2006). Anxiety disorders during pregnancy and the postpartum period: A systematic review. *Journal of Clinical Psychiatry*, 67(8), 1285-1298. doi: 10.4088/JCP.V67n0818
- Ruiz, R. J., & Avant, K. C. (2005). Effects of maternal prenatal stress on infant outcomes: a synthesis of the literature. *Advances in Nursing Science*, 28(4), 345-355. doi: 10.1097/00012272-200510000-00006
- Rychik, J., Donaghue, D. D., Levy, S., Fajardo, C., Combs, J., Zhang, X., Szwast, A., & Diamond, G. S. (2013). Maternal psychological stress after prenatal diagnosis of congenital heart disease. *The Journal of Pediatrics*, 162(2), 302-307. doi: 10.1016/j.jpeds.2012.07.023
- Sklansky, M., Tang, A., Levy, D., Grossfeld, P., Kashani, I., Shaughnessy, R., & Rothman, A. (2002). Maternal psychological impact of fetal echocardiography. *Journal of the American Society of Echocardiography*, 15(2), 159-166. doi: 10.1067/mjc.2002.116310
- Smith, M. V., Shao, L., Howell, H., Lin, H., & Yonkers, K. A. (2011). Perinatal depression and birth outcomes in a Healthy Start project. *Maternal and Child Health Journal*, 15(3), 401-409. doi: 10.1007/s10995-010-0595-6
- Solberg, O., Gronning Dale, M. T., Vollrath, M. E., Holmstrom, H., Eskedal, L. T., & Landolt, M. A. (2011). Long-term symptoms of depression and anxiety in mothers of infants with congenital heart defects. *Journal of Pediatric Psychology*, 36(2), 179-187. doi: 10.1093/jpepsy/jsq054

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

- Solberg, O., Gronning Dale, M. T., Vollrath, M. E., Landolt, H., Holstrom, H., & Eskedal, L. T. (2012). Trajectories of maternal mental health: A prospective study of mothers of infants with congenital heart defects from pregnancy to 36 months postpartum. *Journal of Pediatric Psychology*, 37(6), 687-696. doi: 10.1093/jpepsy/jss044
- Sommerseth, E., & Sundby, J. (2010). Women's experiences when ultrasound examinations give unexpected findings in the second trimester. *Women and Birth*, 23(3), 111-116. doi: 10.1016/j.wombi.2010.01.001
- Speilberger, C. D., Gorsuch, R. L., Lushene, R., Vagg, P. R., & Jacobs, G. A. (1983). *Manual for the State-Trait Anxiety Inventory*. Palo Alto, CA: Consulting Psychologists Press.
- Springett, A., Budd, J., Draper, E. S., Kurinczuk, J. J., Medina, J., Rankin, J., Rounding, C., Tucker, D., Wellesley, D., Wreyford, B., & Morris, J. K. *Congenital Anomaly Statistics 2012: England and Wales*. London: British Isles Network of Congenital Anomaly Registers. 2014.
- Van Den Bergh, B. R., Mennes, M., Mulder, E. J. H., & Glover, V. (2005). Antenatal maternal anxiety and stress and the neurobehavioral development of the fetus and child: Links and possible mechanisms. A review. *Neuroscience and Biobehavioral Reviews*, 29(2), 237-258. doi: 10.1016/j.neurobiorev.2004.10.007
- Van Horn, M., DeMaso, D. R., Gonzalez-Heydrich, J., & Erikson, J. D. (2001). Illness-related concerns of mothers of children with congenital heart disease. *Journal of the American Academy of Child and Adolescent Psychiatry*, 40(7), 847-854. doi: 10.1097/00004583-200107000-00020
- Verreault, N., Da Costa, D., Marchand, A., Ireland, K., Dritsa, M., & Khalife, S. (2014). Rates and risk factors associated with depressive symptoms during pregnancy and with postpartum onset. *Journal of Psychosomatic Obstetrics and Gynaecology*, 35(3), 84-91. doi: 10.3109/0167482X.2014.947953
- Weiss, D. S., & Marmar, C. R. (1996). The Impact of Event Scale – Revised. In J. Wilson & T. M., Keane (Eds.), *Assessing Psychological Trauma and PTSD* (pp.399-411). New York: Guilford.
- Williams, J. W., Plassman, B. L., Burke, J., Holsinger, T., Benjamin, S. (2010). Preventing Alzheimer's Disease and Cognitive Decline. Evidence Report/Technology Assessment No. 193. (Prepared by the Duke Evidence-based Practice Center under Contract No. HHS 290-2007-10066-I.) AHRQ Publication No. 10-E005. Rockville, MD: Agency for Healthcare Research and Quality.
- World Health Organisation (2008). *Improving maternal mental health*. Millennium development goal 5 – improving maternal health. Retrieved from http://www.who.int/mental_health/prevention/suicide/Perinatal_depression_mmh_fin_al.pdf?ua=1
- Yanikkerem, E., Ay, S., Mutlu, S., & Goker, A. (2013). Antenatal depression: prevalence and risk factors in a hospital based Turkish sample. *Journal of The Pakistan Medical Association*, 63(4), 472-477.

- Zhu, M., Liang, H., Feng, J., Wang, Z., Wang, W., & Zhou, Y. (2016). Effectiveness of medical self-experience counselling in pregnant women after echocardiographic detection of a suspected fetal heart anomaly: A longitudinal study. *Journal of Obstetrics and Gynaecology Research*, 42(10), 1236-1244. doi: 10.1111/jog.13
- Zigmond, A. S., & Snaith, R. P. (1983). The hospital anxiety and depression scale. *Acta Psychiat Scand*, 67, 361-370.

CHAPTER 2

EMPIRICAL PAPER

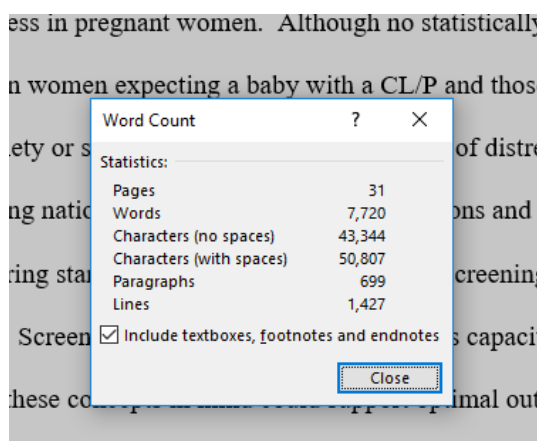
Expecting a Baby with a Cleft: Maternal Mental Health, Antenatal Attachment, and the Role of Mindfulness and Self-Compassion¹

Lisa Halpin² (BSc, MSc), Julie Van Vuuren² (BSc, DClínPsy), Ross White² (BSc, PhD, DClínPsy), Zoe Edwards³ (MA, ClínPsyD), and Rachel Mumford³ (BSc, ClínPsyD)

²University of Liverpool, UK.

³Alder Hey Children's National Health Service Foundation Trust, UK.

Word count: 7,720



Author Note

Thanks are extended to Dr Peter Taylor for his support and guidance in the early stages of this study and Caroline Cunningham (Research Midwife) for supporting this study and facilitating data collection at Liverpool Women's Hospital.

¹Manuscript intended for submission to The Journal of Prenatal and Perinatal Psychology and Health (word limit: 2000-8000; Appendix A)

Abstract

The impact of a cleft lip and/or palate (CL/P) diagnosis on maternal mental health, and associations with antenatal attachment (AA), mindfulness and self-compassion (SC) was explored. Pregnant women expecting a baby with a CL/P ($n = 27$) and pregnant women with no such diagnosis ($n = 61$) were recruited. No significant differences in maternal mental health were found between groups. Significant correlations were found in each group between SC, mindfulness, and psychological distress variables. The small sample size in the CL/P group limits generalisability of the findings, however recommendations are made for antenatal services, and future research.

Keywords: cleft, pregnancy, mental health, antenatal attachment, mindfulness, self-compassion.

Introduction

Cleft lip and/or palate (CL/P) is one of the most common craniofacial abnormalities (Cleft Lip and Palate Association [CLAPA]). The aetiology of CL/P is thought to be multifactorial, with a complex interaction between genetic and environmental factors (Dixon, Marazita, Beaty & Murray, 2011). It is caused by a failure of lip and/or palate elements to fuse adequately within the first trimester of pregnancy (Berkowitz, 2013). Incidence rates vary worldwide based on ethnicity and geographical location but in the United Kingdom (UK), approximately one in every 700 babies are born with a CL/P (CLAPA). In 2015, 44% of babies born with a CL/P in the UK were diagnosed prenatally (Medina, Fitzsimmons, Copley, Deacon & van der Meulen, 2016).

The type of cleft diagnosed will determine the care and treatment required. For example, a cleft lip often requires less surgical intervention than a bilateral cleft lip and palate, which has a greater impact on facial appearance and feeding, particularly in new-born babies (Chetpakdeechit, Hallberg, Hagberg & Mohlin, 2009; Rumsey & Stock, 2013). However, regardless of diagnoses, a multi-disciplinary team of surgeons, nurses, dentists, speech and language therapists and clinical psychologists, are often involved in the child's care long-term.

Psychological Response to Cleft Diagnosis

Often, prenatal diagnosis is viewed as more favourable than postnatal diagnosis (Berggren, Hansson, Uvemark, Svensson & Becker, 2012), as it allows parents time to prepare practically and psychologically (Aspinall, 2002). Nevertheless, prenatal CL/P diagnosis can be associated with emotional distress and multiple concerns amongst parents (Kuttenberger, Ohmer, & Polska, 2010). Research has identified parental concerns relating to their child's appearance, feeding difficulties, the process and outcomes of surgery, uncertainty regarding their child's future, and self-blame for the development of the CL/P

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

(Hedrick, 2005; Hsieh, Chao & Shiao, 2013; Johansson & Ringsberg, 2004; Reid, Kilpatrick & Reilly, 2006; Skari et al., 2006; Titapant & Chuenwattana, 2015). Furthermore, additional ethical and moral dilemmas might be experienced if abortion following diagnosis is considered (Blumenfield, Blumenfield & Bronshtein, 1999; Johnson & Sandy, 2003).

Prenatal CL/P diagnosis has also been associated with a sense of loss and grieving for the ideal child (Titapant & Chuenwattana, 2015) and feelings of shock, helplessness, denial, anger, fear and guilt (Bradbury & Hewison, 1994; Jones, 2002). Furthermore, prenatal diagnosis of non-lethal congenital anomalies, have also been associated with increased levels of anxiety, depression and stress in pregnant women (Hedrick, 2005; Kemp, Davenport & Pernet, 1998; Lalor, Devane & Begley, 2007). This is important when one considers the detrimental impact and influence that maternal anxiety and/or depression can have on the mother (e.g. postnatal depression), foetal development (Van den Bergh, Mulder, Mennes & Glover, 2005), pre-term birth and/or low birth-weight (Ding et al., 2014), and long term implications for a child's physical and mental wellbeing (Bauer, Knapp & Parsonage, 2015).

Attachment and Cleft

Psychological distress (e.g. depression, anxiety, stress) during pregnancy can also impact on the maternal-foetal bond (Alhusen, 2008; Lindgren, 2001; Rubertsson, Pallant, Sydsjo, Haines & Hildingsson, 2015). Schmidt, Seehagen, Vocks, Schneider and Teismann (2016) recently found that rumination and worrying within the first four months of pregnancy were significant predictors of impairments in antenatal attachment in later pregnancy, and statistically significant levels of depression and anxiety. There has been much debate regarding the conceptualisation and definition of maternal-foetal relationships, and if the term attachment can be applied to the prenatal period at all (see Walsh, 2010). Indeed, Bowlby (1958) defined attachment as a reciprocal behavioural process *prompted by an infant to*

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

ensure survival, however observations of mothers and their new-borns, and grief reactions of mothers following miscarriage, has inspired interest in the concept of prenatal bonding (see Brandon, Pitts, Denton, Stringer & Evans, 2009).

John Condon (1993) introduced the term antenatal attachment (AA) to capture how a mother seeks closeness to her foetus and strives to identify and meet its needs, and to protect it. Defined simply, AA is “the emotional tie or bond, which normally develops between the pregnant parent and her unborn child” (Condon & Corkindale, 1997, p.359). AA involves cognitive representations of the foetus and how the mother behaves in relation to this. This includes actions and behaviours such as maintaining a healthy diet, buying items for the baby, talking to and caressing the pregnancy ‘bump’ (Sailsbury, Law, LaGrase & Lester, 2003).

Research has found that the postnatal maternal-infant relationship and attachment is significantly reduced when the child has a visible congenital anomaly (Boztepe, Ay, Kerimoglu Yildiz & Cinar, 2016). Studies focussing on maternal gaze have illustrated how the degree of mutual gaze between a mother and infant can predict relationship quality (Britton, Gronwaldt & Britton, 2001). Yet, mothers of babies with a cleft lip focus less on the mouth region, have fewer positive responses to their infants and have reduced gaze generally (De Pascalis et al., 2017; De Pascalis et al., 2015; Murray et al., 2008; Parsons et al., 2011). Likewise, infants with a cleft lip are rated by adults as less ‘cute’ than infants without a cleft (Rayson et al., 2016) and are more likely to have mothers who are emotionally distant from them than mothers of infants without a cleft (Despars, Peter, Borghini and Hohlfeld, 2011). These implications for mother-infant interactions and responsiveness extend further still, with infants with a CL/P classified as having a secure attachment style at 12 months of age being more avoidant upon reunion with their mothers than securely attached children without a CL/P (Habersaat et al., 2013).

Mindfulness and Self-Compassion

Given the above findings, understanding factors that might promote psychological well-being and AA in pregnant women expecting a baby with a CL/P is therefore important. Mindfulness and self-compassion (SC) have been associated with improved mental health (Barnard & Curry, 2011), and positive psychological outcomes (e.g. happiness) (Hollis-Walker & Colosimo, 2011; Neff, Kirkpatrick & Rude, 2007). Although mindfulness and SC are often used in conjunction (e.g. Mindful Self-Compassion program; Germer & Neff, 2013) they are distinct concepts.

Mindfulness relates to acknowledging difficult thoughts and feelings and responding to them with openness and curiosity. Kabat-Zinn (1994) defines mindfulness as “paying attention in a particular way, on purpose, in the present moment and non-judgementally” (p.4). Mindfulness is considered as both a dispositional trait, and a skill that requires cultivation through meditation (Bodhi, 2000; Brown & Ryan, 2003, Brown, Ryan & Creswell, 2007). Mindful individuals are thought to be more able to effectively regulate and manage their emotions through enhanced metacognitive awareness (Masicampo & Baumeister, 2007). Mindfulness has received substantial support for its association with increased positive emotions and reduced psychological distress (Brown et al., 2007; Khoury, Sharma, Rush & Fournier, 2015; Masicampo & Baumesiter, 2007). A recent systematic review specifically highlighted the effectiveness of mindfulness interventions for reducing anxiety, worry, and depression in pregnant women (Matvienko-Silkar, Lee, Murphy & Murphy, 2016).

As mindfulness advocates a curious attitude towards difficult thoughts and feelings, SC advocates responding to such experiences with kindness and sympathy to offer comfort to oneself (Germer & Neff, 2013; Smeets, Neff, Alberts & Peters, 2014). SC has been defined

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

as “...being caring and compassionate toward oneself in the face of hardship or perceived inadequacy...having the right amount of distance from one’s emotions so that they are fully experienced while being approached with mindful objectivity” (Neff et al., 2007, p. 140).

High levels of depression and anxiety have been associated with low levels of SC in pregnant women (Felder, Lemon, Shea, Kripke & Dimidjian, 2016), however interventions to enhance SC in pregnant women are associated with reductions in psychological distress (Dunn, Hanich, Roberts & Powrie, 2012), indicating that SC may be a protective factor against psychological distress (Xavier et al., 2016). Importantly, higher levels of SC have also been related to greater maternal-fetal attachment (Cohen, 2010; Mohamadirizi & Kordi, 2016).

Aims and Hypotheses

This study aims to contribute to the evidence base by examining in more detail differences between pregnant women expecting a baby with a CL/P and women without such a diagnosis on a range of factors. Specifically, psychological distress (e.g. depression, anxiety and stress), AA, mindfulness and SC. Associations between these variables in each group of pregnant women, will also be explored. It was hypothesised that:

1. Pregnant women expecting a baby with a CL/P, relative to those who are not, will report statistically significant higher levels of depression, anxiety and stress

In each group of pregnant women:

2. Higher levels of depression, anxiety and stress will be associated with lower levels of AA
3. Higher levels of mindfulness will be associated with lower depression, anxiety and stress, and higher levels of AA

4. Higher levels of SC will be associated with lower levels of depression, anxiety and stress, and higher levels of AA
5. Psychological distress and AA will be significantly predicted by a regression model that includes CL/P status, mindfulness and SC.

Method

Eligibility Criteria

Participants who had received their 20-week anomaly scan (a routine ultrasound scan offered to all pregnant women in the UK at approximately 20 weeks' gestation to screen for anomalies) were eligible to participate. Due to associations with reduced AA and to reduce potential further confounding variables, participants were excluded from the study if they met the following criteria: under 18 years of age (Rowe, Wynter, Steele, Fisher & Quinlivan, 2013), IVF treatment for current pregnancy (McMahon, Ungerer, Beaurepaine, Tennant & Sanders, 1997; Stanton & Golombok, 1993), twin pregnancies or diagnosis of a foetal anomaly other than CL/P. Inability to comprehend and communicate in the English language was also exclusion criteria.

Participants

A total of 126 people were recruited via opportunistic and self-selected sampling between March 2016 and May 2017. Following inspection of the data, 38 participants met exclusion criteria for the following reasons: aged under 18 years ($n = 2$); not had 20-week scan ($n = 7$); twin pregnancy ($n = 2$); received IVF for current pregnancy ($n = 5$); foetus diagnosed with another anomaly ($n = 10$); no measures completed ($n = 12$). The final sample comprised 88 pregnant women. Table 1 displays clinical/demographic characteristics and chi-square results for each group.

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

Table 1

Participant Characteristics, Clinical Variables and χ^2 results

Variable	Cleft Group (<i>n</i> = 27)	Non-Cleft Group (<i>n</i> = 61)	χ^2	<i>df</i>	<i>p</i>
Maternal age – years	29.8 (4.8) [23-42]	31.9 (4.9) [21-43]			
≤ 29	59.3%	27.9%	7.91	2	.019
30-34	18.5%	37.7%			
35+	18.5%	29.5%			
Missing	3.7% (<i>n</i> = 1)	4.9% (<i>n</i> = 3)			
Gestation – weeks	30.7 (6.3) [20-42]	29.55 (5.5) [20-39]			
≤ 27	29.6%	36.1%	.45	2	.840
28-33	37%	34.4%			
34+	33%	27.9%			
Missing	0.0%	1.6% (<i>n</i> = 1)			
Sex of baby:					
Male	74.1%	36.1%	11.06	1	.003
Female	14.8%	23%			
Known, but not stated	0.0%	13.1%			
Not known	11.1%	26.2%			
Missing	0.0%	1.6% (<i>n</i> = 1)			
3D/4D scan completion:					
Yes	70.4%	23.0%	15.56 ^a	1	.000
No	29.6%	75.4%			
Missing	0.0%	1.6% (<i>n</i> = 1)			
Miscarriage history:					
Yes	33.3%	29.5%	.00 ^a	1	.805
No	66.7%	68.9%			
Missing	0.0%	1.6% (<i>n</i> = 1)			
Other children:					
Yes	48.1%	52.4%	.65 ^a	2	.779
No	51.9%	45.9%			
Missing	0.0%	1.6% (<i>n</i> = 1)			
Maternal anomalies:					
Yes	0.0%	8.2%	1.10 ^a	1	.178
No	100%	90.2%			
Missing	0.0%	1.6% (<i>n</i> = 1)			

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

Ethnicity:						
White English/Welsh/Scottish/Northern	85.2%	86.9%				
Irish	3.7%	0.0%				
White Irish	3.7%	4.9%				
Any other white background	3.7%	0.0%				
Mixed/multiple: White and black	3.7%	0.0%	.60	1		.662
Caribbean	0.0%	3.3%				
Asian/Asian British: Chinese	0.0%	1.6%				
Any other mixed/multiple ethnic background	0.0%	3.3%				
Black/African/Black British: Caribbean						
Any other ethnic group not specified						
Marital status:						
Single	11.1%	13.1%				
Married	59.3%	47.5%		1		1.000
Divorced	3.7%	0.0%	.05			
Cohabiting	25.9%	37.7%				
Civil Partnership	0.0%	1.6% (<i>n</i> = 1)				
Employed:						
Yes	85.2%	75.4%	1.06	1		.404
No	14.8%	24.5%				
Qualification level:						
GCSE	14.8%	13.1%				
NVQ	7.4%	16.4%				
A/AS-Level	18.5%	9.8%	.02	1		1.000
Degree	48.1%	39.3%				
Masters	11.1%	16.4%				
Doctorate/PhD	0.0%	4.9% (<i>n</i> = 3)				
Cleft type:						
CL/P	70.4%	0.0%				
Cleft Lip	11.1%	0.0%	N/A	N/A		N/A
Not known	18.5%	0.0%				

Note. Data are shown as mean, (SD) and [range]. N/A = Not applicable.

^aYates' Correction for Continuity used

Materials

A demographics questionnaire (Appendix F) collected information relating to participants age, ethnicity, marital status, employment status, qualifications, gestational age, sex of baby, cleft type and attendance at cleft support groups (if applicable), 3D/4D scan completion, details regarding any other children and their ages, history of miscarriage, and if the participants themselves were born with any congenital anomalies. Participants also completed four self-report measures (Appendix G), detailed below:

Maternal Antenatal Attachment Scale (MAAS; Condon, 1993).

The MAAS measures maternal attachment to the foetus and comprises 19 items with five answer choices that vary between questions, divided into two subscales: quality of attachment and intensity of attachment. Quality of attachment (10 items) includes statements such as: ‘when I first see my baby after the birth I expect I will feel...’ followed by 5 responses. Intensity of attachment (9 items) includes statements such as: ‘over the past two weeks, I have thought about, or been preoccupied with the baby inside me’. Ten items are reverse scored. A global attachment score is calculated by summing all the items. Good internal consistency was reported by Condon (1993) for the global attachment score (Cronbach’s $\alpha=.82$). Alpha values for the quality subscale have been reported as ranging between .65 and .80, and between .66 to .77 for the intensity subscale (Mako & Deak, 2014; Van Bussel, Spitz & Demyttenaere, 2010). This study found acceptable coefficients also (global = .72; attachment quality = .75; attachment intensity = .66).

Depression Anxiety Stress Scales (DASS-21; Lovibond & Lovibond, 1995).

The DASS-21 measures the severity of a range of core symptoms associated with depression, anxiety and stress. It comprises 21 items evenly distributed throughout the scales and are rated on a scale ranging from 0 (*never*) to 3 (*almost always*). Items associated with the three scales include: ‘I felt down-hearted and blue’ (depression), ‘I felt scared without any good reason’ (anxiety), and ‘I found it hard to wind down’ (stress). The final scale scores are multiplied by two, and can be categorised (based on population mean) as normal, mild, moderate, severe and extremely severe. Good internal consistency has been reported for the DASS-21 scales (depression: $\alpha=.88$; anxiety: $\alpha=.82$; stress: $\alpha=.90$) (Henry & Crawford, 2005). Acceptable coefficients were found in the present study (depression: $\alpha=.87$; anxiety: $\alpha=.69$; stress: $\alpha=.82$).

Five Facet Mindfulness Questionnaire (FFMQ; Baer, Smith, Hopkins, Kreitemeyer & Toney, 2006).

The FFMQ is a 39-item questionnaire that assesses five conceptual facets of mindfulness: observing (e.g. ‘I pay attention to how my emotions affect my thoughts and behaviour’), describing (e.g. ‘I’m good at finding words to describe my feelings’), acting with awareness (e.g. ‘I am easily distracted’), non-judging of inner experience (e.g. ‘I disapprove of myself when I have irrational ideas’) and non-reactivity to inner experience (e.g. ‘I watch my feelings without getting lost in them’). Statements are rated on a scale ranging from 1 (*never or very rarely true*) to 5 (*very often or always true*). Nineteen items are reverse scored. Good internal consistency has been reported for all facets (observe: $\alpha = .83$; describe: $\alpha = .91$; acting with awareness $\alpha = .87$; non-judge $\alpha = .87$; non-react $\alpha = .75$) (Baer et al., 2006). This was maintained in the present study (observe $\alpha = .66$; describe: $\alpha = .89$; acting with awareness $\alpha = .88$; non-judge $\alpha = .85$; non-react $\alpha = .77$).

Although the individual subscales of the FFMQ are usually examined, a ‘global mindfulness’ score can also be computed by summing the subscale total scores (Williams, Dalgleish, Karl & Kuyken, 2014). Williams, Dalgleish, Karl and Kuyken (2014) argue that in a community sample that do not practice meditation, the ‘observe’ subscale should be omitted and the FFMQ should be considered a ‘four-factor’ model of mindfulness, not a ‘five-factor’ model. For the purposes of this research, a global mindfulness score was used, and calculated without the ‘observe’ subscale. Acceptable internal consistency was found for the summed scale ($\alpha = .89$).

Self-compassion Scale (SCS; Neff, 2003).

The SCS comprises 26-items containing six subscales: self-kindness (e.g. ‘I’m kind to myself when I’m experiencing suffering’) self-judgement (e.g. ‘when times are really

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

difficult, I tend to be tough on myself'), common humanity (e.g. 'I try to see my failings as part of the human condition'), isolation (e.g. 'When I fail at something that's important to me, I tend to feel alone in my failure'), mindfulness (e.g. 'When something upsets me I try to keep my emotions in balance'), and over-identification (e.g. 'When something upsets me I get carried away with my feelings'). Responses range from 1 (*almost never*) to 5 (*almost always*). Using a total score is recommended (Neff, Whittaker & Karl, 2017) and calculated by reverse coding negatively worded items, generating subscale means and summing. Good internal consistency has been reported for the total scale ($\alpha = .92$). The present study also found acceptable coefficients (self-kindness $\alpha = .85$; self-judgement $\alpha = .83$; common humanity $\alpha = .79$; isolation $\alpha = .84$; mindfulness $\alpha = .72$; overidentified $\alpha = .81$; total $\alpha = .94$).

Procedure

This study obtained ethical approval from the National Health Service (NHS) Health Research Authority (HRA) (Appendix H). To recruit pregnant women expecting a baby with a cleft, advertisements were placed on the CLAPA (a CL/P charity offering support to individuals and families) website and Facebook page. Posters and flyers advertising the research were distributed to clinical nurse specialists (CNSs) and clinical psychologists working across a regional NHS CL/P service based at Alder Hey Children's Hospital, Royal Manchester Children's Hospital, and Leeds General Infirmary. Advertisements contained information relating to the nature of the study, and a link to a secure online version of the study if they wished to access further information and complete it online. Questionnaire packs were also distributed to CNSs who agreed to distribute them during routine antenatal appointments if they felt it was appropriate to do so.

Pregnant women attending routine antenatal clinics (e.g. no suspected foetal anomalies) at Liverpool Women's Hospital were approached in person regarding the study.

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

Following an overview of the research and an opportunity to read the participant information sheet (Appendix I), potential participants were invited to take part and complete the questionnaires either in the clinic waiting area, at home and return the questionnaires using the pre-paid envelope, or online using the link provided. Consent forms were completed by all participants before commencing the study (Appendix J). Completion of the questionnaires took approximately 20 minutes. In recognition of participant's time and effort, they were given the opportunity to enter a prize draw to win one of three £50 high street shopping vouchers and to receive a summary of the study findings upon completion of the research.

Figure 1 provides an illustration of the flow of participants through the study.

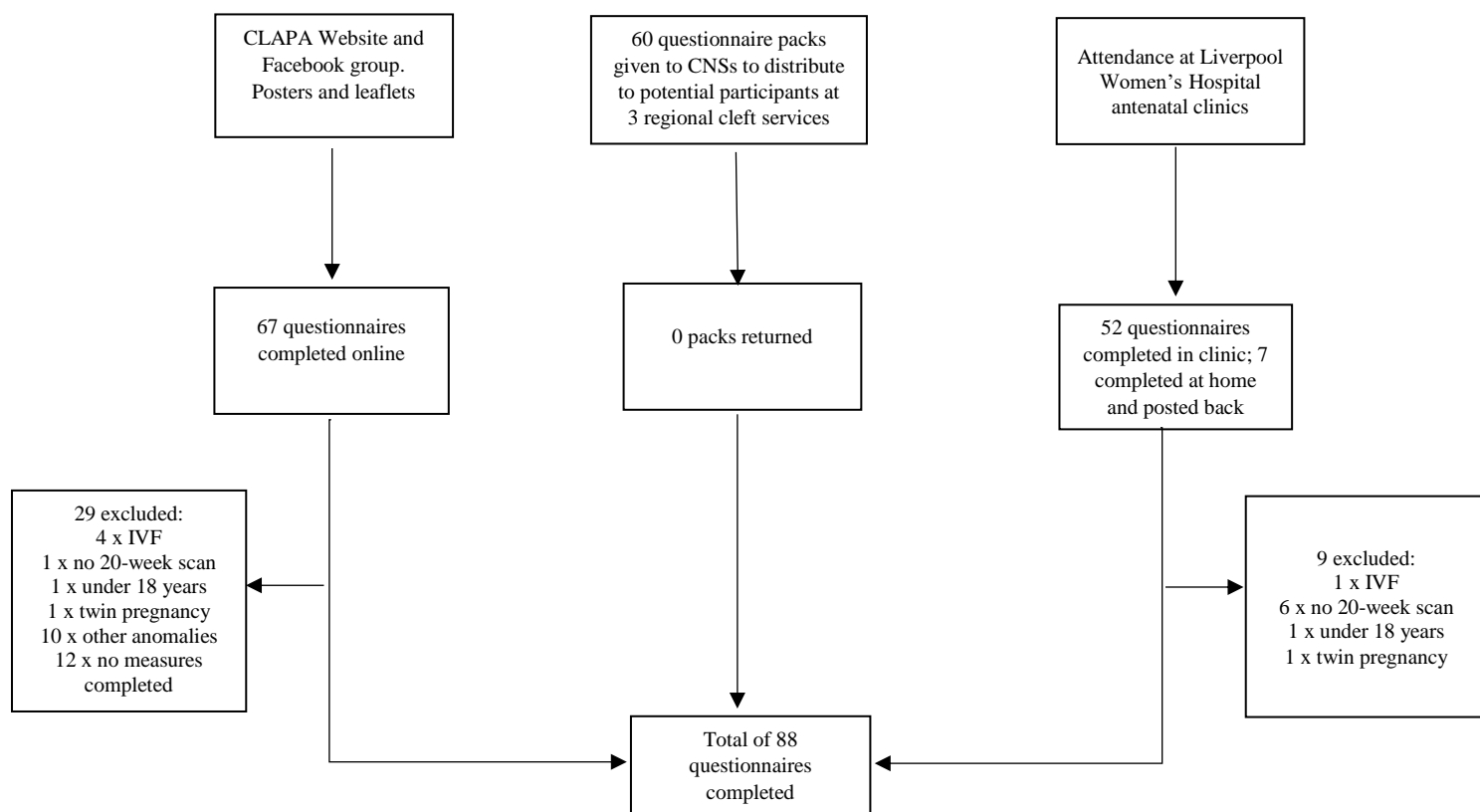


Figure 1. Flow of participants through the study

Power Calculation

A priori power calculation using G*Power (version 3.1; Faul, Erdfelder, Lang & Buchner, 2007) suggested an approximate sample size of 85 (with equal n in both groups) to detect a medium sized effect ($f^2=.15$) with power of 80% when employing the $\alpha=.05$ criterion of statistical significance in multiple regression analyses. Depression, anxiety and stress were the main outcome variables, in addition to AA. CL/P status, mindfulness and SC were the predictor variables. Due to uneven group sizes in the final sample, and undertaking a different analysis (MANCOVA) a post-hoc power calculation using G*Power was conducted. Using the above criteria, this revealed sufficient power (98%).

Statistical Analyses

All analyses were conducted using SPSS version 24. Normality was assessed for each of the variables and grouped based on cleft/non-cleft diagnosis. Inspection of skewness and kurtosis values, plots and the Kolmogorov-Smirnov statistic revealed positively skewed data for the depression, anxiety and stress variables for both groups. Positively skewed data on variables such as these, in non-clinical populations is perhaps expected. To minimise the impact of outliers ($n = 6$) their scores were changed to a less extreme value by using the next highest value in the distribution and making it one unit larger (Tabachnick & Fidell, 2013). Normality was checked again following these procedures and the data was still skewed. For analysis involving these skewed variables, non-parametric alternatives were used.

The association between demographic/clinical characteristics and cleft status was assessed using chi-square analyses. The continuous variables maternal age and gestational age were collapsed into categories. Maternal age (years) was categorised as: ≤ 29 ; 30-34; 35+ and gestational age (weeks) was categorised as: ≤ 27 ; 28-33; 34+. Independent samples t -tests were used to explore differences between the groups on AA, mindfulness and SC. Mann-

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

Whitney U tests were used to explore differences between groups on total DASS-21 scores. Chi-square analyses were used to explore differences between groups on DASS-21 clinical significance categories. Pearson's correlation and Spearman's rank order correlations were used to test associations in each group on depression, anxiety, stress, AA (global; quality; intensity), mindfulness and SC. Although the intention was to conduct a multiple regression analysis, the uneven group numbers made this unviable. MANCOVA was used instead with cleft status as the independent variable and psychological distress, AA, mindfulness and SC as the dependant variables. Maternal age, sex of baby and 3D/4D scan completion were the covariates, based on findings from the initial chi-square analyses. Where applicable, Cohen's (1988) effect size criteria were used throughout.

Missing Data

Overall, less than 15% of the data was missing for the outcome variables. Results of Little's MCAR test indicated that data was missing completely at random ($\chi^2(811) = 677.05$, $p = 1.00$). Missing data was addressed using person-mean imputation for six cases where three items or less were missing for the same measure. Cases with four or more items missing on a measure were excluded from analysis using the pairwise deletion method (Pallant, 2013). This equated to $n = 9$ in the cleft group and $n = 5$ in the non-cleft group.

Results

Tests of Differences

A chi-square test for independence was used to explore relationships between pregnant women on clinical/demographic characteristics. Statistically significant associations were found between maternal age and group, sex of baby and group, and completion of 3D/4D scan and group. No significant associations were found between the

A series of independent samples *t*-tests were conducted on all dependant variables between both groups. No significant results were found. Table 2 displays group differences on the study variables. Due to positively skewed data on the DASS-21, Mann-Whitney U Tests were used to explore differences between pregnant women in the cleft and non-cleft group on their self-reported scores on the DASS-21. This test revealed no significant differences between the groups on depression ($U = 515.00, z = -.087, p = .93, r = -.9.98$); anxiety ($U = 520.00, z = -.025, p = .98, r = -2.87$); or stress ($U = 475.00, z = -.577, p = .56, r = -0.07$).

Group Differences for all Study Variables

	Cleft Group			Non-Cleft Group						
Variable	<i>M</i>	<i>SD</i>	<i>n</i>	<i>M</i>	<i>SD</i>	<i>n</i>	<i>df</i>	<i>t</i>	<i>p</i>	Cohen's <i>d</i>
Depression	5.83	5.51	18	6.21	6.06	58	74	-0.23	.816	0.07
Anxiety	6.83	3.79	18	7.71	6.18	58	74	-0.57	.573	0.17
Stress	13.28	6.32	18	14.62	7.77	58	74	-0.67	.507	0.19
Mindfulness	96.26	14.39	19	101.80	16.16	56	73	-1.33	.189	0.36
Self-compassion	3.18	.55	23	3.01	.72	60	81	1.03	.307	0.27
Global Attachment	80.30	5.11	27	79.62	6.80	60	85	0.46	.644	0.11
Attachment Quality	45.11	3.66	27	46.07	3.63	61	86	-1.14	.260	0.26
Attachment Intensity	30.33	4.03	27	28.74	4.69	61	86	1.53	.129	0.36

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

Clinical cut-offs for depression, anxiety and stress were also inspected. Chi-square analysis using Fishers Exact Test statistic revealed no statistically significant differences between the groups on the categories for depression ($p = .75$), anxiety ($p = .32$) or stress ($p = .80$). Table 3 displays frequencies, medians and ranges for the DASS-21 for the cleft and non-cleft group.

Table 3

Clinical Cut-off Frequencies, Medians and Ranges for DASS-21 Scores for the Cleft Group and Non-Cleft Group

Variable (cut-off scores)	Cleft Group ($n = 18$)			Non-Cleft Group ($n = 58$)		
	n (%)	Md^a	Range	n (%)	Md^a	Range
Depression		3	0-17		4	0-29
Normal (0-9)	12 (66.7)			43 (74.1)		
Mild (10-13)	4 (22.2)			10 (17.2)		
Moderate (14-20)	2 (11.1)			3 (5.2)		
Extremely severe (28+)	0 (0.0)			2 (3.4)		
Anxiety		6	0-13		6	0-23
Normal (0-7)	10 (55.6)			34 (58.6)		
Mild (8-9)	2 (11.1)			4 (6.9)		
Moderate (10-14)	6 (33.3)			10 (17.2)		
Severe (15-19)	0 (0.0)			7 (12.1)		
Extremely severe (20+)	0 (0.0)			3 (5.2)		
Stress		13	4-25		14	0-34
Normal (0-14)	12 (66.7)			34 (58.6)		
Mild (15-18)	3 (16.7)			9 (15.5)		
Moderate (19-25)	3 (16.7)			9 (15.5)		
Severe (26-33)	0 (0.0)			5 (8.6)		
Extremely severe (34+)	0 (0.0)			1 (1.7)		

Note: ^a Median for total scores.

Tests of Associations

Pearson product-moment correlation and Spearman rank order correlation was conducted to examine the relationship between mindfulness, self-compassion, AA and psychological distress for both groups. Correlation matrices, means and standard deviations are displayed in Table 4 and Table 5 for both groups respectively.

Table 4

Correlation Matrix of Study Variables: Cleft Group

Variable	<i>n</i>	1	2	3	4	5	6	7	8
1 Self-compassion	23	-							
2 Mindfulness	19	.77**	-						
3 Global Attachment	27	.08	.11	-					
4 Depression	18	-.38	-.42	-.02	-				
5 Anxiety	18	-.07	-.29	-.11	.68**	-			
6 Stress	18	-.62**	-.70**	.20	.71**	.47*	-		
7 Attachment Intensity	27	-.29	-.05	.71**	.29	.23	.41	-	
8 Attachment Quality	27	.44*	.27	.58**	-.35	-.46	-.33	-.16	-

* $p < .05$, ** $p < .01$

Table 5

Correlation Matrix of Study Variables: Non-cleft Group

Variable	<i>n</i>	1	2	3	4	5	6	7	8
1 Self-compassion	60	-							
2 Mindfulness	56	.77**	-						
3 Global Attachment	60	.15	.11	-					
4 Depression	58	-.51**	-.65**	-.00	-				
5 Anxiety	58	-.39**	-.58**	.10	.67**	-			
6 Stress	58	-.60**	-.76**	-.14	.63**	.66**	-		
7 Attachment Intensity	61	-.08	-.08	.81**	.15	.30*	.10	-	
8 Attachment Quality	61	.32*	.32*	.71**	-.26	-.25	-.42**	.19	-

* $p < .05$, ** $p < .01$

Controlling for Covariates

A one-way between groups multivariate analysis of covariance (MANCOVA) was performed to see if differences existed between groups when controlling for certain variables. Eight dependant variables were used: mindfulness, self-compassion, global attachment, attachment quality, attachment intensity, depression, anxiety and stress. The independent variable was foetal diagnosis (e.g. cleft/non-cleft). Maternal age, sex of baby and 3D/4D scan completion were used as covariates in this analysis.

The MANOVA/MANCOVA function in SPSS was used for the analysis with the sequential adjustment for nonorthogonality. Preliminary assumption testing confirmed that there were no serious violations of normality, linearity, homogeneity of variance-covariance and multicollinearity for all variables except anxiety. The assumption of equality was

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

violated for anxiety, indicated by a significant Levene's Test statistic ($p = .03$). An adjusted alpha of .01 was therefore used for this variable (Tabachnick & Fidell, 2013).

There was a statistically significant difference between the groups of pregnant women on a linear combination of the dependant variables, $F(8, 58) = 3.36, p = .003$; Wilks' Lambda = .68; with a large effect size (partial $\eta^2 = .32$). There was also a statistically significant effect of the covariate age on a linear combination of the dependant variables, $F(8, 58) = 3.71, p = .001$; Wilks' Lambda = .66; with a large effect size (partial $\eta^2 = .34$). The covariates sex of baby and 3D/4D scan were not significantly related to the dependant variables among the groups $F(8, 58) = 1.11, p = .37$ and $F(8, 58) = .76, p = .64$, respectively.

The dependant variables were investigated separately using a Bonferroni adjusted alpha level of .006 and .001 (for the adjusted anxiety alpha) to reduce the chance of a Type I error (Pallant, 2013). No statistically significant differences were found between the groups and their scores on the dependant variables. There was a statistically significant relationship between the covariate age and attachment intensity when controlling for the independent variable (i.e. group), $F(1, 65) = 14.01, p = .000$, partial $\eta^2 = .18$.

To investigate these significant findings further, the univariate test statistics were inspected. This revealed no statistically significant differences between the groups and the dependant variables once the effect of the covariate had been statistically removed. Table 6 presents adjusted mean scores, standard errors, confidence intervals, and univariate statistics for the study variables for both groups.

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

Table 6

MANCOVA Adjusted Means, Standard Errors, Confidence Intervals and Univariate Test Statistics for Outcome Variables

Note. AA = Antenatal attachment; SC = Self-compassion.

Cleft Group					Non-Cleft Group				Univariate		
(n = 18)					(n = 52)						
Variable	M	SE	95% CI		M	SE	95% CI		F(1,65)	p	η ²
			Lower	Upper			Lower	Upper			
Global AA	78.78	1.64	75.50	82.06	79.77	.92	77.93	81.61	.26	.612	.004
AA Quality	44.90	.92	43.07	46.73	46.00	.51	44.97	47.02	1.01	.318	.015
AA Intensity	29.11	1.05	27.01	31.21	29.10	.59	27.92	30.27	.00	.992	.000
Mindfulness	96.68	4.11	88.47	104.90	100.98	2.31	96.37	105.58	.77	.383	.012
SC	3.29	.17	2.95	3.64	2.93	.10	2.74	3.12	3.13	.082	.046
Depression	5.16	1.49	2.18	8.12	6.18	.84	4.51	7.84	.33	.568	.005
Anxiety	5.58	1.40	2.79	8.37	7.99	.78	6.43	9.56	2.10	.152	.031
Stress	12.03	1.90	8.23	15.82	15.16	1.06	13.04	17.29	1.94	.169	.029

Discussion

Prenatal diagnosis of congenital anomalies can impact on maternal mental health, and consequently foetal development and AA. Therefore, it is imperative to identify factors that might help to ameliorate psychological distress and enhance AA. Mindfulness and SC are identified as two such factors in the literature. This study aimed to identify the prevalence of psychological distress amongst pregnant women with a foetal CL/P diagnosis in comparison to pregnant women without such a diagnosis. This study also aimed to explore associations between psychological distress, AA, mindfulness and SC.

The hypothesis that depression, anxiety and stress would be statistically significantly higher in the cleft group was not supported. The hypothesis that higher levels of depression, anxiety and stress will be associated with lower levels of AA was partially supported in the non-cleft group. The hypothesis that higher levels of mindfulness will be associated with lower levels of depression, anxiety and stress was supported in the non-cleft group and partially supported in the cleft group. Mindfulness was only significantly associated with attachment quality in the non-cleft group. The hypothesis that higher levels of SC will be associated with lower levels of depression, anxiety and stress, was supported in the non-cleft group and partially supported in the cleft group. SC was significantly associated with AA quality in both the cleft and non-cleft group. Maternal age was significantly associated with AA intensity.

The finding that psychological distress was not significantly greater in the cleft group contradicts previous research (e.g. Hedrick, 2005; Kemp et al., 1998; Lalor et al., 2007; Titapant & Chuenwattana, 2015). However, this finding does perhaps support research conducted by Aspinall (2002) and Berggren et al. (2012) who found that prenatal diagnosis gave parents time to psychologically prepare and adjust for the birth of their baby.

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

Furthermore, women in the cleft group might have received *extra* practical and emotional support from CNSs and CLAPA, which might explain the lack of statistically significant differences between the groups. Additionally, on average, there was a 10-week period between women having their 20-week scan and completion of the measures. As the concepts measured can be considered states, not traits, perhaps it could be expected that at the point of completing the measures, distress may have reduced from what it was initially.

Although most participants in the cleft group had scores within the clinically ‘normal’ range for depression, anxiety and stress, some participants were experiencing clinically significant levels in the mild to moderate range. Interestingly, only participants in the non-cleft group reported experiencing clinically significant levels of depression, anxiety and stress in the ‘severe’ or ‘extremely severe’ range. These findings support the need for antenatal services to offer mental health screening to all women attending routine antenatal appointments.

A significant negative association between attachment quality and stress was found in the non-cleft group, which supports previous research (Alhusen, 2008; Rubertsson et al., 2015). Although causation cannot be inferred, perhaps increased stress, which is related to difficulties relaxing, feeling irritable and being easily upset, reduces attachment *quality*, which is associated with experiences of closeness and tenderness to the foetus (Condon & Corkindale, 1997). Furthermore, anxiety and attachment intensity were found to have a positive association, which contradicts previous research (Schmidt et al., 2016). However, as attachment intensity relates to the extent to which the baby is embedded in the woman’s emotional life (Condon & Corkindale, 1997), maybe women who are more likely to feel anxious generally, experience increased preoccupation, or intensity, towards their unborn baby.

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

Perhaps significant associations between psychological distress and AA in the cleft group were not found due to increased completion of the 3D/4D scan. Seventy percent of women in the cleft group had a 3D/4D scan compared to 30% of women in the non-cleft group. It could be hypothesised that 3D/4D scans defend against psychological distress and therefore AA is protected. However, the effectiveness of 3D/4D ultrasound beyond that of 2D ultrasound for reducing maternal anxiety (Leung et al., 2006), for example, and enhancing maternal-foetal bonding (De Jong-Pleij et al., 2013; Ji et al., 2005) remains inconclusive.

Significant negative associations between mindfulness and psychological distress, and SC and psychological distress were found across both groups, supporting previous research (Barnard & Curry, 2011; Neff et al., 2007; Xavier et al., 2016). However, only mindfulness and stress, and SC and stress were negatively associated in the cleft group, in contrast to associations with mindfulness and SC (respectively), and stress, depression and anxiety in the non-cleft group. Again, although causality cannot be inferred, increases in mindfulness and SC (respectively) may have been associated with decreases in psychological distress (Germer & Neff, 2013).

Mindfulness was also significantly positively associated with attachment quality in the non-cleft group. It could be that increased mindfulness is associated with increased attachment quality as mindfulness promotes non-judgemental present moment awareness, which in turn might promote closeness and tenderness towards the unborn baby. Perhaps mindfulness was not associated with attachment intensity as this involves a preoccupation with the unborn baby, and individuals who exhibit increased mindfulness, are less likely to spend time in a preoccupied state. In both groups, however, SC and attachment quality were significantly positively associated. This supports the findings of Cohen (2010) and Mohamadiriz and Kordi (2016) who found that SC was related to increased maternal-foetal attachment. Again, although causation cannot be inferred, perhaps the kinder pregnant

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

women are towards themselves (e.g. SC), the more closeness and tenderness they can perhaps feel towards their unborn baby (e.g. attachment quality).

The significant association between maternal age and attachment intensity requires further investigation. Researchers have commented on associations between parental age and AA (Damato, 2005; van Bakel, Maas, Vreeswijk & Vingerhoets, 2013) and so it appears that this is worthy of further exploration.

Several strengths and limitations are noted from this study. There is no published research examining the impact of prenatal cleft diagnosis on maternal mental health, AA, and the influence of mindfulness and SC. This study therefore offers a preliminary exploration of this novel area. Mothers of children with clefts and CNSs at regional cleft services were consulted during the early stages of the research and encouraged to offer their suggestions. This helped to influence and develop a meaningful study.

The small sample size in the cleft group has undoubtedly impacted on the findings as it was anticipated that significant differences would be found between the two groups in terms of mental health difficulties. Specifically, that women in the cleft group would report higher levels of psychological distress. Although the non-cleft group reported a slightly higher incidence of clinical levels of distress, this is possibly because of the uneven group sizes. The results of the correlation analysis and subsequent conclusions are also limited for this reason. Indeed, increasing the number of participants in the cleft group would be advantageous to make more robust conclusions. However previous studies have highlighted the difficulties associated with recruiting pregnant women to sensitive research about foetal diagnoses (Lalor & Begley, 2006; Paton, Wood, Bor & Nitsun, 1999; Statham, Solomou & Green, 2001). Barriers to recruiting women expecting a baby with a cleft from cleft antenatal support groups, significantly affected the ability to achieve a larger sample

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

size, and perhaps identify significant differences between the groups on distress. Increasing sample size would allow for more advanced data analyses such as regression, mediation and/or moderation, which would help to explore the influence of mindfulness and SC in more detail. Matching the two groups might have also been advantageous, however, this may have further restricted the size of the cleft group.

In addition to the small sample size, missing data was also an issue in the cleft group, where in some cases only one of the four questionnaires were completed. As the cleft group was hypothesised to experience greater psychological distress, perhaps lack of questionnaire completion reflects those participants who were experiencing significant psychological distress. Perhaps if face-to-face recruitment had been possible, this would have improved completion of the measures. Failure to randomise the order of the measures presented to participants, might have also affected why some participants did not proceed beyond the first questionnaire, which was the MAAS.

Although the missing values analyses showed that there was no pattern to the missing data, the MAAS does contain some questions that could be potentially upsetting to participants (e.g. “Some pregnant women sometimes get so irritated by the baby inside them that they feel like they want to hurt or punish it”). To reduce the effect of missing data, transformation was considered. However, it was decided that it would not be appropriate in this study, especially because the small cleft sample size already decreased the ability to generalise findings, and transformation may have affected this further. Transformation is also a heavily debated topic within the research literature and is often criticised (Tabachnick & Fidell, 2013).

The measures chosen in this study also have implications for the results found. The DASS was chosen as it has been used in research with pregnant women before (Xavier, et al.,

2016), however perhaps a pregnancy specific measure, such as the Pregnancy Related Anxiety Questionnaire-Revised 2 (PRAQ-R2; Huizink, et al., 2016) or the Tilburg Pregnancy Distress Scale (TPDS; Pop, et al., 2011) would have been more appropriate. Pregnancy specific measures often omit somatic complaints (e.g. fatigue; dry mouth; over-reacting) that are usually observed during pregnancy due to hormone changes, and are also a feature of depression, anxiety and stress. As the DASS includes numerous physiological symptoms and items relating to difficulties relaxing, for example, this might have impacted on the findings in this study, which further limits conclusions drawn.

Similarly, the use of the FFMQ, rather than a questionnaire assessing *dispositional* mindfulness might have further affected results. Although the FFMQ is a well-validated measure of mindfulness, its facets largely comprise of components from the Kentucky Inventory of Mindfulness Skills (KIMS; Baer, Smith & Allen, 2004). This is an issue in this study because the KIMS was developed to measure the cultivation of mindfulness skills based on psychological therapies that include mindfulness training such as mindfulness based stress reduction, for example. Collecting data on participant's mindfulness practice or knowledge of mindfulness as a concept would have been beneficial. Furthermore, the language used in the FFMQ might not have been familiar to individuals who do not practice mindfulness or are not aware of what it entails. For example, language such as noticing thoughts and bodily sensations; watching and stepping back from feelings, and noticing visual elements in nature might have felt alien to some participants.

Future research would benefit from enhancing participant numbers in the cleft group. This could be improved by involving a range of international cleft charities and support forums, as opposed to just CLAPA. Recruiting women expecting a baby with a CL/P face-to-face also warrants further exploration. Perhaps spending more time with the CNSs discussing and confirming recruitment strategies might be beneficial. Whilst some missing

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

data is expected, strategies that help to facilitate complete participation is worth exploring. In this study, participants who did not complete all the measures, were generally recruited online. It appears that some participants started the questionnaires, but perhaps became fatigued, got distracted, or simply changed their mind about taking part in the study. Randomising the order, and selecting shorter versions of questionnaires, where appropriate, could also enhance completion. It might have also been helpful to send reminders to participants who had started, but not completed the questionnaires online. However, this is not without ethical implications, and would require careful consideration.

Future studies may also consider employing additional control groups including pregnant women who have received different foetal diagnoses. This would enable an exploration of differences in AA, psychological distress and factors associated with enhancing psychological wellbeing based on diagnosis. Longitudinal designs capturing data at various time points throughout pregnancy would also be advantageous, especially as prior research has shown changes in attachment and psychological distress throughout pregnancy (Cannella, 2005; Lee et al., 2007). Therefore, identifying the most appropriate time to offer support and intervention is also worthy of further exploration. Collecting data prior to conception of pregnancy, where feasible, would also allow for important pre-post comparisons of psychological distress.

Future studies might also wish to consider demographic variables in more depth. For example, in this study almost 60% of participants in the cleft group were aged 29 years or younger. Tailoring recruitment strategies to allow for this demographic might improve engagement in the research. For example, using a range of social media platforms rather than just Facebook. Additionally, examining in more depth the impact of demographic and clinical factors such as being a first-time mother, previous miscarriages, gestational age and

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

variables such as psychological distress, AA, mindfulness and SC would be worthy of exploration due to potential theoretical associations.

SC and attachment quality appear to promote one another. Investigating whether there is a link between them could also be a focus of future research. Understanding AA and its components is also worthy of further exploration. In this study, global attachment was not related to any of the study variables illustrating that perhaps focusing on total AA scores is insufficient, and examination of individual components is warranted, as demonstrated in this study.

Mindfulness, SC and aspects of AA appear to be important factors for antenatal services to consider when working with pregnant women. Although exploratory in nature, this study has highlighted some interesting associations both within, and between these concepts and psychological distress. Furthermore, the results suggest that regardless of prenatal diagnosis, a proportion of pregnant women experience clinically significant levels of distress.

Health professionals working in antenatal services are well placed to support the identification of maternal mental health difficulties. Although the National Institute for Clinical Health and Excellence (NICE) offer generic guidelines recommending that perinatal mental health difficulties are routinely screened for (NICE, 2014), they are not mandatory and variability regarding implementation (or lack of) will exist between services (Glover, 2014). Standardised, routine mental health screening for all pregnant women is therefore recommended. Alternatively, given the significant associations found in this study between mindfulness, SC and psychological distress, perhaps screening for reduced SC and mindfulness might support identification of pregnant women who might benefit from additional emotional support. This approach to screening might reduce the stigma associated

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

with mental health difficulties and might be viewed as a more acceptable, and positive approach to mental health screening by pregnant women. Further research is warranted, however.

Fortunately, in their Five Year Forward View publication, NHS England (2014) acknowledged perinatal mental health as an area requiring more funding and service development. Overall, £365m has been allocated to specialist perinatal mental health services to improve access and treatment to 30,000 more women by 2021. In the latest Five Year Forward View update, NHS England (2017) announced the development of 20 new or expanded perinatal mental health teams. Additionally, 29 New Model of Care Programme vanguard sites across the UK have also been established which enable local organisations to get involved in co-designing and establishing new care models (NHS England, 2017).

Alongside other health professionals, clinical psychologists (CP) can support the development of perinatal mental health services by contributing to funding applications and facilitating the implementation of mental health screening and more comprehensive assessments where appropriate. CP's can also offer teaching and training sessions to staff working with pregnant women to promote evidence-based practice, in addition to co-facilitating, or supervising interventions (Inc. group and/or individual) aimed at reducing psychological distress in pregnant women. Although this study did not find any significant differences between pregnant women expecting a baby with a CL/P and pregnant women without such a diagnosis, previous research highlights that foetal diagnosis can exacerbate or contribute to reduced maternal mental health. Clinicians should therefore be particularly aware of this when supporting pregnant women following such a diagnosis.

Conclusion

Despite its limitations, this study offers initial exploratory findings that mindfulness, SC and components of AA warrant further exploration in relation to their association with psychological distress in pregnant women. Although no statistically significant differences were found between women expecting a baby with a CL/P and those without such a diagnosis on depression, anxiety or stress, clinically significant levels of distress were evident in both groups. Considering national guidelines and key NHS visions and policies for perinatal mental health, offering standardised, routine mental health screening to all pregnant women seems appropriate. Screening and assessing for mindfulness capacity and SC, and developing interventions with these concepts in mind could support optimal outcomes for pregnant women. This warrants further exploration and so additional research is therefore required.

References

- Alhusen, J. L. (2008). A literature update on maternal-fetal attachment. *Journal of Obstetric, Gynaecologic and Neonatal Nursing*, 37(3), 315-328. doi: 10.1111/j.1552-6909.2008.00241.x
- Aspinall, C. L. (2002). Dealing with the prenatal diagnosis of clefting: A parent's perspective. *Cleft Palate-Craniofacial Journal*, 39(2), 183-187.
- Baer, R. A., Smith, G. T., & Allen, K. B. (2004). Assessment of mindfulness by self-report: The Kentucky Inventory of Mindfulness Skills. *Assessment*, 11, 191-206. doi: 10.1177/1073191104268029
- Baer, R. A., Smith, G. T., Hopkins, J., Kreitemeyer, J., & Toney, L. (2006). Using self-report assessment methods to explore facets of mindfulness. *Assessment*, 13, 27-45.
- Barnard, L. K., & Curry, J. F. (2011). Self-compassion: Conceptualisations, correlates and interventions. *Review of General Psychology*, 15(4), 289-303. doi: 10.1037/a0025754
- Bauer, A., Knapp, M., & Parsonage, M. (2015). Lifetime costs of perinatal anxiety and depression. *Journal of Affective Disorders*, 192, 83-90. doi: 10.1016/j.jad.2015.12.005
- Berggren, H., Hansson, E., Uvemark, A., Svensson, H., & Becker, M. (2012). Prenatal compared with postnatal cleft diagnosis: what do parents think? *Journal of Plastic Hand Surgery*, 46, 235-241. Doi: 10.3109/2000656x.2012.698416
- Berkowitz, S. (2013). Facial and Palatal Growth: In Berkowitz, S. *Cleft Lip and Palate: Diagnosis and Management* (pp.45-58). Retrieved from https://link-springer-com.liverpool.idm.oclc.org/chapter/10.1007%2F978-3-642-30770-6_3
- Blumenfield, Z., Blumenfield, I., & Bronshtein, M. (1999). The early prenatal diagnosis of cleft lip and the decision-making process. *Cleft Palate Craniofacial Journal*, 36(6), 105-107.
- Bodhi, B. (2000). *A comprehensive manual of Abhidhamma: Pali text, translation and explanatory guide*. Onalaska: Buddhist Publication Society Pariyatti Editions. Retrieved from http://www.saraniya.com/books/meditation/Bhikkhu_Bodhi-Comprehensive_Manual_of_Abhidhamma.pdf
- Bowlby, J. (1958). The nature of the child's tie to his mother. *International Journal of Psycho-Analysis*, 39, 350-373.
- Boztepe, H., Ay, A., Kerimoglu Yildiz, G., & Cinar, S. (2016). Does the visibility of a congenital anomaly affect maternal-infant attachment levels? *Journal of Specialist Pediatric Nursing*, 21(4). doi: 10.1111/jspn.12157
- Bradbury, E. T., & Hewison, J. (1994). Early prenatal adjustment to visible congenital disfigurement. *Child: Care, Health & Development*, 20(4), 251-266. doi: 10.1111/j.1365-2214.1994.tb00388.x
- Brandon, A. R., Pitts, S., Denton, W. H., Stringer, A.C., & Evans, H. M. (2009). A history of the theory of prenatal attachment. *Journal of Prenatal and Perinatal Psychology and Health*, 23(4), 201-222.

- Britton, H. L., Gronwaldt, V., & Britton, J. R. (2001). Maternal postpartum behaviours and mother-infant relationship during the first year of life. *The Journal of Pediatrics*, 138(6), 905-909. doi: 10.1067/mpd.2001.113358
- Brown, K. W., & Ryan, R. M. (2003). The benefits of being present: Mindfulness and its role in psychological wellbeing. *Journal of Personality and Social Psychology*, 84, 822-848. doi: 10.1037/0022-3514.84.4.822
- Brown, K. W., Ryan, R. M., & Creswell, J. D. (2007). Mindfulness: Theoretical foundations and evidence for its salutary effects. *Psychological Inquiry*, 18(4), 211-237.
- Cannella, B. L. (2005). Maternal-fetal attachment: An integrative review. *Journal of Advanced Nursing*, 50, 60-68.
- Chetpakdeechit, W., Hallberg, U., Hagberg, C., & Mohlin, B. (2009). Social life aspects of young adults with cleft lip and palate: grounded theory approach. *Acta Odontologica Scandinavica*, 67(2), 122-128. doi: 10.1080/00016350902720888
- Cleft Lip and Palate Association (CLAPA). What is a cleft lip and palate? Retrieved from <https://www.clapa.com/what-is-cleft-lip-palate/>
- Cohen, J. W. (1988). *Statistical power analysis for the behavioural sciences* (2nd ed.). Hillsdale, NJ: Lawrence Erlbaum Associates.
- Cohen, J. (2010). Mindfulness and self-compassion in the transition to motherhood: A prospective study of postnatal mood and attachment. [PhD dissertation], Colombia University.
- Condon, J. (1993). The assessment of antenatal emotional attachment: Development of a questionnaire instrument. *British Journal of Medical Psychology*, 66(2), 167-183. doi: 10.1111/j.2044-8341.1993.tb01739.x
- Condon, J., & Corkindale, C. (1997). The correlates of antenatal attachment in pregnant women. *British Journal of Medical Psychology*, 70(4), 359-372. doi: 10.1111/j.2044-8341.1997.tb01912.x
- de Jong-Pleij, E. A., Ribbert, L. S., Pistorius, L. R., Tromp, E., Mulder, E. J., & Bilard, C. M. (2013). Three-dimensional ultrasound and maternal bonding, a third trimester study and review. *Prenatal diagnosis*, 33(1), 81-88. doi: 10.1002/pd.4013
- Demato, E. (2005). Predictors of prenatal attachment in mothers of twins. *Journal of Obstetric Gynaecologic & Neonatal Nursing*, 33(4), 436-445. doi: 10.1177/0884217504266894
- De Pascalis, L., Bozicevic, L., Hawkins, L., Kkeli, N., Bicknell, S., & Murray, L. (2015). The effect of cleft lip and palate on maternal gaze patterns during early interactions, and their role in maternal responses to early communicative behaviours. *Journal of Reproductive and Infant Psychology*, 33(3), E17-E18.
- De Pascalis, L., Kkeli, N., Chakrabarti, B., Dalton, L., Vaillancourt, K., Rayson, H.,...Murray, L. (2017). Maternal gaze to the infant face: Effects of infant age and

- facial configuration during mother-infant engagement in the first nine weeks. *Infant Behaviour and Development*, 46, 91-99. doi: 10.1016/j.infbeh.2016.12.003
- Despars, J., Peter, C., Borghini, A., & Hohlfeld, J. (2011). Impact of a cleft lip and/or palate on maternal stress and attachment representations. *The Cleft Palate-Craniofacial Journal*, 48(4), 419-24. doi: 10.1597/08-190
- Ding, X., Wu, Y., Xu, S., Zhu, R., Jia, X., Zhang, S., ... Tao, F. (2014). Maternal anxiety during pregnancy and adverse birth outcomes: A systematic review and meta-analysis of prospective cohort studies. *Journal of Affective Disorders*, 159, 103-110. doi: 10.1016/j.jad.2014.02.027
- Dixon, M. J., Marazita, M. L., Beaty, T. H., & Murray, J. C. (2011). Cleft lip and palate: synthesizing genetic and environmental influences. *Nature Reviews Genetics*, 12(3), 167-178. doi: 10.1038/nrg2933
- Dunn, C., Hanich, E., Roberts, R., & Powrie, R. (2012). Mindful pregnancy and childbirth: effects of a mindfulness based intervention on women's psychological distress and well-being in the perinatal period. *Archives of Women's Mental Health*, 15, 139-143. doi: 10.1007/s00737-012-0264-4
- Faul, F., Erdfelder, E., Lang, A. G., & Buchner, A. (2007). G*power 3: a flexible statistical power analysis program for the social, behavioural, and biomedical sciences. *Behavioural Research Methods*, 39(2), 175-191.
- Felder, J., Lemon, E., Shea, K., Kripke, K., & Dimidjian, S. (2016). Role of self-compassion in psychological wellbeing among perinatal women. *Archives of Women's Mental Health*, 19(4), 687-690. doi: 10.1007/s00737-016-0628-2
- Germer, C. K., & Neff, K. D. (2013). Self-compassion in clinical practice. *Journal of Clinical Psychology: In session*, 69(8), 1-12. doi: 10.1002/jclp.2202
- Glover, V. (2014). Maternal depression, anxiety and stress during pregnancy and child outcome; what needs to be done. *Best Practice and Research Clinical Obstetrics and Gynaecology*, 28, 25-35. doi: 10.1016/j.bpobgyn.2013.08.017
- Habersaat, S., Monnier, M., Peter, C., Bolomey, L., Borghini, A., Despars, J., ... Hohlfeld, J. (2013). Early mother-child interaction and later quality of attachment in infants with an orofacial cleft compared to infants without cleft. *The Cleft Palate-Craniofacial Journal*, 50(6), 704-712. doi: 10.1597/12-094.1
- Hedrick, J. (2005). The lived experience of pregnancy while carrying a child with a known, nonlethal congenital anomaly. *Journal of Obstetric, Gynecologic and Neonatal Nursing*, 34(6), 732-740. doi: 10.1177/0884217505282020
- Henry, J. D., & Crawford, J. R. (2005). The short-form version of the depression anxiety stress scales (DASS-21): Construct validity and normative data in a large non-clinical sample. *British Journal of Clinical Psychology*, 44, 227-239. doi: 10.1348/014466505x29657

- Hollis-Walker, L., & Colosimo, K. (2011). Mindfulness, self-compassion and happiness in non-meditators: A theoretical and empirical examination. *Personality and Individual Differences*, 50, 222-227. doi: 10.1016/j.paid.2010.09.033
- Hsieh, Y., Chao, Y.Y., & Shiao, J. S. (2013). A qualitative study of psychosocial factors affecting expecting mothers who choose to continue a cleft lip and/or palate pregnancy to term. *The Journal of Nursing Research*, 21(1), 1-8. doi: 10.1097/jnr.0b013e3182828dbd
- Huizink, A. C., Delfortiere, M. J., Scheinin, N. M., Tolvanen, M., Karlsson, L., & Karlsson, H. (2016). Adaption of pregnancy anxiety questionnaire-revised for all pregnant women regardless of parity: PRAQ-R2. *Archive of Womens Mental Health*, 19, 125-132. doi: 10.1007/s00737.015.0531.2
- Ji, E. K., Pretorius, D. H., Newton, R., Uyans, K., Hull, A. D., Hollenbach, K., & Nelson, T. R. (2005). Effects of ultrasound on maternal-fetal bonding: a comparison of two- and three-dimensional imaging. *Ultrasound, Obstetrics & Gynaecology*, 25, 473-477. doi: 10.1002/uog.1896
- Johansson, B., & Ringsberg, K. C. (2004). Parents' experiences of having a child with cleft lip and palate. *Journal of Advanced Nursing*, 47(2), 165-173. doi: 10.1111/j.1365-2648.2004.03075.x
- Johnson, N., & Sandy, J. R. (2003). Prenatal diagnosis of cleft lip and palate. *Cleft Palate Craniofacial Journal*, 40, 186-189.
- Jones, M. C. (2002). Prenatal diagnosis of cleft lip and palate: detection rates, accuracy of ultrasonography, associated anomalies, and strategies for counselling. *Cleft Palate - Craniofacial Journal*, 39(2), 169-173.
- Kabat-Zinn, J. (1994). *Wherever you go, there you are: Mindfulness meditation in everyday life*. New York, NY: Hyperion.
- Kemp, J., Davenport, M., & Pernet, A. (1998). Antenatally diagnosed surgical anomalies: The psychological effect of parental antenatal counselling. *Journal of Pediatric Surgery*, 33(9), 1376-1379. doi: 10.1016/soo22-3468(98)90011-2
- Khoury, B., Sharma, M., Rush, S. E., & Fournier, C. (2015). Mindfulness-based stress reduction for healthy individuals: A meta-analysis. *Journal of Psychosomatic Research*, 78(6), 519-528. doi: 10.1016/j.j.psychores.2015.03.009
- Kuttenberger, J., Ohmer, J. N., & Polska, E. (2010). Initial counselling for cleft lip and palate: parent's evaluation, needs and expectations. *International Journal of Oral and Maxillofacial Surgery*, 39(3), 214-220. doi: 10.1016/j.ijom.2009.12.013
- Lalor, J., & Begley, C. (2006). Fetal anomaly screening: what do women want to know? *Issues and Innovations in Nursing Practice*, 11-19, doi: 10.1111/j.1365-2648.2006.03884.x

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

- Lalor, J., Devane, D., & Begley, C.M. (2007). Unexpected diagnosis of fetal abnormality: women's encounters with caregivers. *Birth*, 34, 80-88. doi: 1111/j.1523-536x.2006.00148.x
- Lee, A. M., Lam, S. K., Sze Mun Lau, S. M., Chong, C. S. Y., Chui, H. W., & Fong, D. Y. T. (2007). Prevalence, course, and risk factors for antenatal anxiety and depression. *Obstetrics and Gynecology*, 110(5), 1102-1112. doi: 10.1097/01.AOG.0000287065.59491.70
- Leung, K.Y., Ngai, C. S. W., Lee, A., Chan, H. Y., Leung, W. C., Lee, C. P., & Tang, M. H. Y. (2006). The effects on maternal anxiety of two-dimensional versus two-plus three-/four-dimensional ultrasound in pregnancies at risk of fetal abnormalities: a randomised study. *Ultrasound, Obstetrics and Gynaecology*, 28, 249-254. doi: 10.1002/uog.2844
- Lindgren, K. (2001). Relationships among maternal-fetal attachment, prenatal depression, and health practices in pregnancy. *Research in Nursing and Health*, 24(3), 203-217. doi: 10.1002/nur.1023
- Lovibond, S. H., & Lovibond, P. F. (1995). *Manual for the Depression Anxiety Stress Scales*. Sydney: Psychology Foundation.
- Mako, H. S., & Deak, A. (2014). Reliability and validity of the Hungarian version of the Maternal Antenatal Attachment Scale. *International Journal of Gynaecological and Obstetrical Research*, 1, 33-44.
- Masicampo, E. J., & Baumeister, R. F. (2007). Relating mindfulness and self-regulatory processes. *Psychological Inquiry*, 18(4), 255-258.
- Matvienko-Silkar, K., Lee, L., Murphy, G., & Murphy, L. (2016). Effects of mindfulness interventions on prenatal well-being: A systematic review. *Psychology and Health*, 31(12), 1415-1434. doi: 10.1080/08870446.2016.1220557
- McMahon, C. A., Ungerer, J. A., Beaurepaine, J., Tennant, C., & Sanders, D. (1997). Anxiety during pregnancy and fetal attachment after in-vitro fertilization conception. *Human Reproduction*, 12(1), 176-182.
- Medina, J., Fitzsimmons, K., Copley, L., Deacon, S., & van der Meulen, J. (2016). *Annual report on cleft lip and/or palate 2016*. Retrieved from <file:///C:/Users/Lisa/Downloads/CRANE%20Annual%20Report%202016%20v1.pdf>
- Mohamadirizi, S., & Kordi, M. (2016). The relationship between multi-dimensional self-compassion and fetal-maternal attachment in the prenatal period in referred women to Mashad Health Center. *Journal of Education and Health Promotion*, 5, 21. doi: 10.4103/2277-9531.184550
- Murray, L., Hentges, F., Hill, J., Karpf, J., Mistry, B., Kreutz, M., & ...Goodacre, T. (2008). The effect of cleft lip and palate and the timing of lip repair on mother-infant

- interactions and infant development. *Journal of Child Psychology and Psychiatry*, 49(2), 115-123. doi: 10.1111/j.1469-7610.2007.01833.x
- Neff, K. D. (2003). Development and validation of a scale to measure self-compassion. *Self and Identity*, 2, 223-250.
- Neff, K. D., Kirkpatrick, K. L., & Rude, S. S. (2007). Self-compassion and adaptive psychological functioning. *Journal of Research in Personality*, 41, 139-154. doi: 10.1016/j.jrp.2006.03.004
- Neff, K. D., Whittaker, T. A., & Karl, A. (2017). Examining the factor structure of the self-compassion scale in four distinct populations: is the use of a total scale score justified? *Journal of Personality Assessment*, 1-12. doi: 10.1080/00223891.2016.1269334
- NHS England. (2014). *Five Year Forward View*. Retrieved from <https://www.england.nhs.uk/wp-content/uploads/2014/10/5yfv-web.pdf>
- NHS England. (2017). *Next steps on the Five Year Forward View*. Retrieved from <https://www.england.nhs.uk/wp-content/uploads/2017/03/NEXT-STEPS-ON-THE-NHS-FIVE-YEAR-FORWARD-VIEW.pdf>
- NICE. (2014). *Antenatal and postnatal mental health: clinical management and service guidance*. Retrieved from <https://www.nice.org.uk/guidance/cg192>
- Pallant, J. (2013). *SPSS Survival Manual: A step by step guide to data analysis using IBM SPSS*. Maidenhead, Berkshire, England: McGraw Hill.
- Parsons, C. E., Young, K. S., Parsons, E., Dean, A., Murray, L., Goodacre, T., & ...Kringelbach, M. L. (2011). The effect of cleft lip on adult's responses to faces: cross-species findings. *Plos One*, 6(10), doi: 10.1371/journal.pone.0025897
- Paton, F., Wood, R., Bor, R., & Nitsun, M. (1999). Grief in miscarriage patients and satisfaction with care in a London hospital. *Journal of Reproductive and Infant Psychology*, 17, 301-305.
- Pop, V. J. M., Pommer, A. M., Pop-Purceanu, M., Wijen, H. A. A., Bergink, V., & Pouwer, F. (2011). Development of the Tilburg Pregnancy Distress Scale: The TPDS. *Pregnancy and Childbirth*, 11, 80. doi: 10.1186/1471.2393.11.80
- Rayson, H., Parsons, C. E., Young, K. S., Goodacre, T. E., Kringelbach, M. L., Bonaitu, J. J., & ...Murray, L. (2016). Effects of infant cleft lip on adult gaze and perceptions of cuteness. *The Cleft Palate-Craniofacial Journal: Official publication of the American Cleft Palate-Craniofacial Association*. doi: 10.1597/16-015
- Reid, J., Kilpatrick, N., & Reilly, S. (2006). A prospective, longitudinal study of feeding skills in a cohort of babies with cleft conditions. *Cleft palate craniofacial journal*, 43(6), 702-709. doi: 10.1597/05-172

- Rowe, H. J., Wynter, K. H., Steele, A., Fisher, J. R., & Quinlivan, J. A. (2013). Growth of maternal-fetal attachment in pregnant adolescents: a prospective cohort study. *Journal of Paediatric and Adolescent Gynecology*, 26(6), 327-333. doi: 10.1016/j.jpag.2013.06.009
- Rubertsson, C., Pallant, J. F., Sydsjo, G., Haines, H. M., & Hildingsson, I. (2015). Maternal depressive symptoms have a negative impact on prenatal attachment – findings from a Swedish community sample. *Journal of Reproductive and Infant Psychology*, 33(2), 153-164. doi: 10.1080/02646838.2014.992009
- Rumsey, N., & Stock, N. M. (2013). Living with a cleft: psychological challenges, support and intervention. In Berkowitz, S (Ed.), *Cleft lip and palate: diagnosis and management* (pp.907-915). Retrieved from https://link.springer-com.liverpool.idm.oclc.org/chapter/10.1007/978-3-642-30770-6_45
- Sailsbury, A., Law, K., LaGrase, L., & Lester, B. (2003). Maternal-fetal attachment. *Journal of the American Medical Association*, 289(13), 1701-1701.
- Schimdt, D., Seehagen, S., Vocks, S., Schneider, S., & Teismann, T. (2016). Predictive importance of antenatal depressive rumination and worrying for maternal-foetal attachment and maternal well-being. *Cognitive Therapy Research*, 40, 565-576. doi: 10.1007/s10608-016-9759-z
- Skari, H., Malt, U. F., Bjornland, K., Egeland, T., Haugen, G., Skreden, M.,... Emblem, R. (2006). Prenatal diagnosis of congenital malformations and parental psychological distress – A prospective longitudinal cohort study. *Prenatal Diagnosis*, 26(11), 1001-1009. doi: 10.1002/pd.1542
- Smeets, E., Neff, K., Alberts, H., & Peters, M. (2014). Meeting suffering with kindness: Effects of a brief self-compassion intervention for female college students. *Journal of Clinical Psychology*, 70(9), 794-807. doi: 10.1002/jclp.22076
- Stanton, F., & Golombok, S. (1993). Maternal-fetal attachment during pregnancy following in vitro fertilization. *Journal of Psychosomatic Obstetric Gynaecology*, 14, 153-158.
- Statham, H., Solomou, W., & Green, J. M. (2001). *When a baby has an abnormality: A study of parent's experiences*. Centre for Family Research, University of Cambridge, Cambridge.
- Tabachnick, B. G., & Fidell, L. S. (2013). *Using Multivariate Statistics*. USA: Pearson.
- Titapant, V., & Chuenwattana, P. (2015). Psychological effects of fetal diagnoses of non-lethal congenital anomalies on the experience of pregnant women during the remainder of their pregnancy. *The Journal of Obstetrics and Gynaecology Research*, 41(1), 77-83. doi: 10.1111/jog.12504
- van Bakel, H. A., Maas, A. B., Vree Swijk, C. M., & Vingerhoets, A. M. (2013). Pictorial representations of attachment: measuring the parent fetus relationship in expectant

- mothers and fathers. *Pregnancy & Childbirth*, 13(1), 1-9. doi: 10.1186/1471-2393-13-138
- Van Bussel, J. C. H., Spitz, B., & Demyttenaere, K. (2010). Reliability and validity of the Dutch version of the maternal antenatal attachment scale. *Archive of Women's Mental Health*, 13, 267-277.
- Van den Bergh, B. R. H., Mulder, E. J. H., Mennes, M., & Glover, V. (2005). Antenatal maternal anxiety and stress and neurobehavioural development of the fetus and child: links and possible mechanisms: A review. *Neuroscience and Behavioral Reviews*, 29, 237-258. doi: 10.1016/j.neubiorev.2004.10.007
- Walsh, J. (2010). Definitions matter: if maternal-fetal relationships are not attachment, what are they? *Archive of Women's Mental Health*, 13(5), 449-451. doi: 10.1007/s00737-010-0152-8
- Williams, M. J., Dalgleish, T., Karl, A., & Kuyken, W. (2014). Examining the factor structures of the five-facet mindfulness questionnaire and the self-compassion scale. *Psychological Assessment*, 26(2), 407-418. doi: 10.1037/a0035566
- Xavier, S., Azevedo, J., Bento, E., Marques, M., Soares, M., Martins, M. J.,...& Pereira, A. T. (2016). Mindfulness, self-compassion and psychological distress in pregnant women. *The Journal of European Psychiatry*, 33, s603. doi: 10.1016/j.eurpsy.2016.01.1775

*Appendix A***Author Guidelines**

The Journal of Prenatal and Perinatal Psychology and Health accepts only original material that is not under consideration by any other publications. Articles should be word-processed and transmitted electronically to the Editor. The Editor reserves the right to edit manuscripts for length, clarity, and conformity with the journal's style. The author should retain his/her copy. American spelling should be used. The paper should be between 2,000 and 8,000 words with a 100-word abstract and at least three keywords. (See further guidelines for submitting a manuscript in the current APA Publication Manual (2009), specifically, "Author Responsibilities" (pp. 228-231).

The journal is interested in publishing theoretical and empirical articles utilizing data gained from clinical work, experimental research, case studies, and self-report.

Among the areas of special interest are:

- Psychological factors that affect conception, pregnancy, labor, delivery and the post-partum period;
- The reciprocal mechanisms of interaction between the pregnant mother and her unborn and sentient child and the mother and her newborn;
- The influence of the family, society, and the environment on the pregnant mother and her unborn child;
- Evidence-based measures that will improve the emotional well-being of mothers, fathers, and newborns;
- The psychological effects of medical technology during conception, pregnancy, labor, and delivery on all parties concerned;
- Methods of prevention and intervention/resolution of prenatal and perinatal traumas with children and adults;
- Interfaces between prenatal and perinatal psychology and medicine, genetics, developmental psychology, anthropology, ethics, and the law.

Illustrations, Figures and Tables

- All illustrations and tables should be included separately from the manuscript (in a separate document) and should be clearly identified in Arabic numerals, showing which is the top of the illustration if this is not obvious. Legends for illustrations, which should be referred to as "Figures," should also be included with the figures. Tables must supplement the text without duplicating it. They should include an appropriate title.

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

- Lettering within an illustration, figure or table should be no smaller than 8 points and no larger than 10 points, and prepared at a resolution sufficient to produce a high-quality image, that is, using computer-generated, professional-level graphic software.
- Illustrations should either be black-and-white glossy photographs or India ink drawings. Color illustrations will only be shown on the digital version. They will be converted to black and white in the print version

APA Style

- Formatting and referencing must follow APA style. References should be limited to work cited in the article, rather than being a bibliography of the topic.

American Psychological Association (2009). *Publication manual of the American Psychological Association* (6th ed.). Washington, DC: Author.

Email submissions to: journal.editor@birthpsychology.com

Appendix B
PRISMA Checklist

Section/topic	#	Checklist item	Reported on page #
TITLE			
Title	1	Identify the report as a systematic review, meta-analysis, or both.	
ABSTRACT			
Structured summary	2	Provide a structured summary including, as applicable: background; objectives; data sources; study eligibility criteria, participants, and interventions; study appraisal and synthesis methods; results; limitations; conclusions and implications of key findings; systematic review registration number.	
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of what is already known.	
Objectives	4	Provide an explicit statement of questions being addressed with reference to participants, interventions, comparisons, outcomes, and study design (PICOS).	
METHODS			
Protocol and registration	5	Indicate if a review protocol exists, if and where it can be accessed (e.g. Web address), and, if applicable, provide registration information including registration number.	
Eligibility criteria	6	Specify study characteristics (e.g. , PICOS, length of follow-up) and report characteristics (e.g., years considered, language, publication status) used as criteria for eligibility, giving rationale.	
Information sources	7	Describe all informaiton of sources (e.g., databases with dates of coverage, contact with study authors to identify additonal studies) in the search and date last searched.	
Search	8	Present full electronic search strategy for at least one database, includng any limits used, such that it could be repeated.	
Study selection	9	State the process for selecting studies (i.e., screening, eligibility, included in systematic review, and, if applicable, included in the meta-analysis.	
Data collection process	10	Describe method of data extraction from reports (e.g. piloted forms, independatly, in duplicate) and any processes for obtaining and confirming data from investigators.	
Data items	11	List and define all variables for which data were sought (e.g., PICOS, funding sources) and any assumptions and simplifications made.	

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

Risk of bias in individual studies	12	Describe methods used for assessing risk of bias of individual studies (including specification of whether this was done at the study or outcome level), and how this information is to be used in any data synthesis.	
Summary measures	13	State the principal summary measures (e.g., risk ratio, difference in means).	
Synthesis of results	14	Describe the methods of handling data and combining results of studies, if done, including measures of consistency (e.g., I ²) for each meta-analysis.	
Risk of bias across studies	15	Specify any assessment of risk of bias that may affect the cumulative evidence (e.g., publication bias, selective reporting within studies)	
Additional analyses	16	Describe methods of additional analyses (e.g., sensitivity or subgroup analyses, meta-regression), if done, indicating which were pre-specified.	
RESULTS			
Study selection	17	Give numbers of studies screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally with a flow diagram.	
Study characteristics	18	For each study, present characteristics for which data were extracted (e.g. study size, PICOS, follow-up period) and provide the citations.	
Risk of bias within studies	19	Present data on risk of bias of each study and, if available, any outcome level assessment (see item 12).	
Results of individual studies	20	For all outcomes considered (benefits or harms), present, for each study: (a) simple summary data for each intervention group (b) effect estimates and confidence intervals, ideally with a forest plot.	
Synthesis of results	21	Present results of each meta-analysis done, including confidence intervals and measures of consistency.	
Risk of bias across studies	22	Present results of any assessment of risk of bias across studies (see item 15).	
Additional analysis	23	Give results of additional analyses, if done (e.g., sensitivity or subgroup analyses, meta-regression [see item 16]).	
DISCUSSION			
Summary of evidence	24	Summarize the main findings including the strength of evidence for each main outcome; consider their relevance to key groups (e.g., healthcare providers, users, and policy makers).	
Limitations	25	Discuss limitations at study and outcome level (e.g., risk of bias), and a review-level (e.g., incomplete retrieval of identified research, reporting bias).	
Conclusions	26	Provide a general interpretation of the results in the context of other evidence, and implications for future research.	
FUNDING			
Funding	27	Describe sources of funding for the systematic review and other support (e.g., supply of data); role of funders for the systematic review.	

Appendix C

Systematic Review Protocol

Title/question: The Psychological Impact of Antenatal Diagnosis of a Foetal Cardiac Anomaly in Pregnant Women: A Systematic Review

Background and Aims: Congenital heart defects (CHDs) are one of the most common congenital anomalies, and occur in approximately 7 per 1000 live births (Dolk, Loane & Garne, 2011). Understandably, this can be an anxiety provoking time for pregnant women due to the ranging severity, and subsequent implications of CHDs (Harris, Franck & Michie, 2012). It is the purpose of this review to explore the psychological implications for women, and factors related to their wellbeing following a prenatal diagnosis of CHD.

Search Strategy and Sources of Information to Identify the Studies

PsycINFO, CINAHL Plus, MEDLINE, and Web of Science databases will be searched. There will be no restrictions for publication period.

Search terms:

“congenital heart disease” OR “congenital heart defect” OR “cardiac anomal*” OR “cardiac disease” OR “heart disease” OR CHD OR “heart anomal*” OR “heart defect” OR “heart disorder”

AND

Preg* OR prenatal OR antenatal OR fetal OR foetal OR fetus OR foetus OR childbearing OR “prenatal diagnosis”

AND

Anx* OR depress* OR stress OR distress OR wellbeing OR “well-being” OR “life satisfaction” OR adjustment OR “mental health” OR happiness OR “quality of life” OR QOL OR “psychological well*”

Selection Criteria (Inclusion and Exclusion Criteria)

Inclusion criteria: the review will consist of studies that 1) include pregnant women (no age restriction) who have received confirmation (or suspected confirmation) that their baby has a cardiac anomaly (any diagnosis); 2) measure the women’s psychological adjustment, which is defined as the experience of anxiety, depression, stress, distress, or impairment in quality of life, life satisfaction and wellbeing 3) are written in English language.

It is expected that some studies will also include data from father’s/partners. If the data can be interpreted separately for mothers, these studies will be included. Additionally, if this is not clear, authors will be contacted and asked to provide results for women only. Published and non-published research will be eligible for inclusion. It is also expected that some studies will collect data longitudinally (e.g. beyond the antenatal period) these studies will be included if the data captured antenatally is available.

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

Where relevant studies employ mixed-methodology (e.g. qualitative and quantitative), and the quantitative data fits the inclusion criteria, these studies will be included.

Exclusion criteria: Studies focussing solely on adoptive/surrogate/foster carers/family members other than mothers will be excluded. Qualitative research will be excluded in addition to non-English language studies. Studies only looking at heart defects detected at birth, will also be excluded.

Data Screening

Search results will be exported into a reference management software. The first stage will involve the removal of duplicates. Next, one researcher will screen the title and abstracts of all studies for suitability. The remaining articles will then be read in full by two independent researchers. Where any disagreement is noted, a third researcher will be consulted. Supplementary searches will be carried out following screening of included articles reference lists. All corresponding authors of included papers will be contacted and asked about any additional published or unpublished studies that meet inclusion criteria. Conference abstracts of potentially eligible research will also be followed up by contacting the authors to identify any associated published or unpublished data.

Study Quality Assessment

All included papers will be screened with a quality assessment tool adapted for the purpose of this review. This tool is adapted from Williams, Plassman, Burke, Holsinger and Benjamin's (2010) assessment tool for observational studies. Each criterion will be graded as "Yes", "No", "Partially", or "Can't tell". Reference: Williams, J., Plassman, B. L., Burke, J., Holsinger, T., & Benjamin, S. (2010). Preventing alzheimer's disease and cognitive decline. Rockville, MD: Agency for Healthcare Research and Quality.

Synthesis

Narrative synthesis will be used to highlight the findings from the review. Tables will be used to display a summary of the included papers.

References

- Dolk, H., Loane, M., Garne, E. (2011). European Surveillance of Congenital Anomalies (EUROCAT) Working Group, Congenital heart defects in Europe: prevalence and perinatal mortality, 2000 to 2005. *Circulation*, 123, 841-849
- Harris, J.M., Franck, L., & Michie, S. (2012). Assessing the psychological effects of prenatal screening tests for maternal and foetal conditions: a systematic review. *Journal of Reproductive and Infant Psychology*, 30 (3), 222-246.

Conflict of Interest

None.

Sources of Financing

The research is being undertaken as part fulfilment of the Doctorate in Clinical Psychology training programme and is funded by the University of Liverpool.

Appendix D

Quality Assessment Tool

Grade each criterion as “Yes,” “No,” “Partially,” or “Can’t tell.” Factors to consider when making an assessment are listed under each criterion. Note that some criteria will only apply to specific types of study.

1. Unbiased selection of the cohort

Factors that help reduce selection bias:

- Inclusion/exclusion criteria
 - Clearly described
 - Antenatal diagnosis reported
- Recruitment strategy
 - Clearly described
 - Relatively free from bias (Attempts at random recruitment are best. selection bias might be introduced, e.g., by recruitment via advertisement)
 - If a comparison group was used, was the sample appropriate, and did the study investigators ensure groups were comparable by matching, etc.

2. Sample size calculated

Factors to consider:

- Did the authors report conducting a power analysis or describe some other basis for determining the adequacy of study group sizes for the primary outcome(s) of interest to us?
- Did the eventual sample size deviate by $\leq 10\%$ of the sample size suggested by the power calculation? (only applicable if power calculation conducted)

3. Adequate description of the cohort?

Factors to consider:

- Age
- Gestational stage
- At what stage of pregnancy diagnosis was received
- Severity of diagnosis given (e.g. mild, moderate, severe)
- History of mental health difficulties
- Do participant’s themselves have a heart defect or other congenital abnormality

4. Validated method for ascertaining psychological adjustment?

Factors to consider:

- Was the method used to ascertain psychological adjustment clearly described? (Details should be sufficient to permit replication in new studies)

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

- Was a valid and reliable measure/s (e.g. standardised, Cronbach Alpha's reported, etc) used to ascertain adjustment? (self-report measures tend to have lower reliability and validity than clinical interview).
- Were these measures implemented consistently across all study participants?
- 5. **Validated method for ascertaining cardiac anomaly?**
Factors to consider:
 - Was the method used to ascertain both confirmed and suspected antenatal diagnosis clearly described? (e.g. consultation with specialist health professionals/multiple ultrasound scans, etc.).
- 6. **Missing data**
Factors to consider:
 - Did missing data from any group exceed 20%?
 - If missing data is present and substantial, were steps taken to minimize bias (e.g., sensitivity analysis or imputation).
- 7. **Analysis controls for confounding data**
Factors to consider for controlled studies:
 - Does the study identify and control for important confounding variables and effect modifiers (moderators)?
 - Factors to consider for studies looking at confounders of psychological adjustment include:
 - Substance use
 - Socio-economic status
 - Ethnicity & Cultural Context
 - Did the study control for likely demographic and clinical confounders? For example, using multiple regression to adjust for demographic or clinical factors likely to be correlated with predictor and outcome?
- 8. **Analytic methods appropriate**
Factors to consider:
 - Was the kind of analysis done appropriate for the kind of outcome data?
 - Dichotomous – logistic regression, survival
 - Categorical – mixed model for categorical outcomes
 - Continuous – ANCOVA, mixed model

Was the number of variables used in the analysis appropriate for the sample size? (The statistical techniques used must be appropriate to the data and consider issues such as controlling for small sample size, clustering, rare outcomes, multiple comparison, and number of covariates for a given sample size.

Appendix E

Data Extraction Form

General information

Citation

Type of publication (e.g. journal article, conference abstract)

Country of origin

Study characteristics

Aim/objectives of the study

Study design

Study inclusion and exclusion criteria

Recruitment procedures used (e.g. details of randomisation, blinding)

Participant characteristics

Age

Gender

Ethnicity

Socio-economic status

Gestational age

Diagnosis

Comorbidities

Intervention and setting

Setting

Description of the intervention(s) and control(s)

Outcome data/results

Statistical techniques used

For each pre-specified outcome:

Whether reported

Definition used in study

Measurement tool or method used

Unit of measurement (if appropriate)

Length of follow-up, number and/or times of follow-up measurements

For all intervention group(s) and control group(s):

Number of participants enrolled

Number of participants included in analysis

Number of withdrawals, exclusions, lost to follow-up

Summary outcome data e.g.

Dichotomous: number of events, number of participants

Continuous: mean and standard deviation

Type of analysis used in study

Results of study analysis e.g.

Dichotomous: odds ratio, risk ratio and confidence intervals, p-value

Continuous: mean difference, confidence intervals

Appendix F

Demographics Questionnaire

1. Age (please state your age in years):**2. Ethnicity (please circle or tick):**

White: English/Welsh/Scottish/Northern Irish	Asian/Asian British: Chinese
White: Irish	Asian/Asian British: Bangladeshi
White: Gypsy or Irish Traveller	Any other Asian/Asian British background
Any other white background	Black/African/Caribbean/Black British: African
Mixed/multiple: White and Black Caribbean	Black/African/Caribbean/Black British: Caribbean
Mixed/multiple: White and Black African	Any other Black/African/Caribbean/Black British background
Mixed/multiple: White and Asian	Arab
Any other mixed/multiple ethnic background	Any other ethnic group not specified
Asian/Asian British: Pakistani	Asian/Asian British: Indian

3. Marital Status (please circle or tick):

Single	Married
Civil Partnership	Divorced
Widowed	Co-habiting

4. Employment Status:

Full-time	Part-time
Unemployed	Retired
Student	Other

5. Qualifications (please circle or tick):

GCSEs	A-levels/AS levels
Degree	Masters
Doctorate/PhD	No formal qualifications
NVQ	

6. How many weeks pregnant are you?

.....

7. Have you had a 3D/4D Scan?

YES/NO (please delete as appropriate)

8. If YES, how many weeks pregnant were you when you had this scan?

.....

9. How many other children do you have?

.....

10. Please select the age categories for your children:

0-5 years
6-10 years
11-18 years

11. Have any of your pregnancies not resulted in a live birth? (e.g. because of a miscarriage)

YES/NO (please delete as appropriate)

12. If you are expecting a baby with a cleft, have you attended an antenatal support group?

YES/NO (please delete as appropriate)

13. Were you born with any anomalies, such as a cleft, or a heart problem?

YES/NO (please delete as appropriate)

If YES, please

specify.....

14. Do you know the sex of your baby? (please delete as appropriate)

Male/Female/Don't know

Appendix G

Measures

MATERNAL ANTENATAL ATTACHMENT SCALE

These questions are about your thoughts and feelings about the developing baby.
Please tick one box only in answer to each question.

- 1) Over the past two weeks I have thought about, or been preoccupied with the baby inside me:
- ☐ Almost all the time
- ☐ Very frequently
- ☐ Frequently
- ☐ Occasionally
- ☐ Not at all
- 2) Over the past two weeks when I have spoken about, or thought about the baby inside me I got emotional feelings which were:
- ☐ Very weak or non-existent
- ☐ Fairly weak
- ☐ In between strong and weak
- ☐ Fairly strong
- ☐ Very strong
- 3) Over the past two weeks my feelings about the baby inside me have been:
- ☐ Very positive
- ☐ Mainly positive
- ☐ Mixed positive and negative
- ☐ Mainly negative
- ☐ Very negative

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

- 4) Over the past two weeks I have had the desire to read about or get information about the developing baby. This desire is:

- ☐ Very weak or non-existent
- ☐ Fairly weak
- ☐ Neither strong nor weak
- ☐ Moderately strong
- ☐ Very strong

- 5) Over the past two weeks I have been trying to picture in my mind what the developing baby actually looks like in my womb:

- ☐ Almost all the time
- ☐ Very frequently
- ☐ Frequently
- ☐ Occasionally
- ☐ Not at all

- 6) Over the past two weeks I think of the developing baby mostly as:

- ☐ A real little person with special characteristics
- ☐ A baby like any other baby
- ☐ A human being
- ☐ A living thing
- ☐ A thing not yet really alive

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

7. Over the past two weeks I have felt that the baby inside me is dependent on me for its well-being:

<input type="checkbox"/>	Totally
<input type="checkbox"/>	A great deal
<input type="checkbox"/>	Moderately
<input type="checkbox"/>	Slightly
<input type="checkbox"/>	Not at all

- 8) Over the past two weeks I have found myself talking to my baby when I am alone

<input type="checkbox"/>	Not at all
<input type="checkbox"/>	Occasionally
<input type="checkbox"/>	Frequently
<input type="checkbox"/>	Very frequently
<input type="checkbox"/>	Almost all the time I am alone

9. Over the past two weeks when I think about (or talk to) my baby inside me, my thoughts:

<input type="checkbox"/>	Are always tender and loving
<input type="checkbox"/>	Are mostly tender and loving
<input type="checkbox"/>	Are a mixture of both tenderness and irritation
<input type="checkbox"/>	Contain a fair bit of irritation
<input type="checkbox"/>	Contain a lot of irritation

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

10. The picture in my mind of what the baby at this stage actually looks like inside the womb is:

- ☐ Very clear
- ☐ Fairly clear
- ☐ Fairly vague
- ☐ Very vague
- ☐ I have no idea at all

11. Over the past two weeks when I think about the baby inside me I get feelings which are:

- ☐ Very sad
- ☐ Moderately sad
- ☐ A mixture of happiness and sadness
- ☐ Moderately happy
- ☐ Very happy

12. Some pregnant women sometimes get so irritated by the baby inside them that they feel like they want to hurt it or punish it:

- ☐ I couldn't imagine I would ever feel like this
- ☐ I could imagine I might sometimes feel like this, but I never actually have
- ☐ I have felt like this once or twice myself
- ☐ I have occasionally felt like this myself
- ☐ I have often felt like this myself

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

13. Over the past two weeks I have felt:

- ☐ Very emotionally distant from my baby
- ☐ Moderately emotionally distant from my baby
- ☐ Not particularly emotionally close to my baby
- ☐ Moderately close emotionally to my baby
- ☐ Very close emotionally to my baby

14. Over the past two weeks I have taken care with what I eat to make sure the baby gets a good diet:

- ☐ Not at all
- ☐ Once or twice when I ate
- ☐ Occasionally when I ate
- ☐ Quite often when I ate
- ☐ Every time I ate

15. When I first see my baby after the birth I expect I will feel:

- ☐ Intense affection
- ☐ Mostly affection
- ☐ Dislike about one or two aspects of the baby
- ☐ Dislike about quite a few aspects of the baby
- ☐ Mostly dislike

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

16. When my baby is born I would like to hold the baby:

- ☐ Immediately
- ☐ After it has been wrapped in a blanket
- ☐ After it has been washed
- ☐ After a few hours for things to settle down
- ☐ The next day

17. Over the past two weeks I have had dreams about the pregnancy or baby:

- ☐ Not at all
- ☐ Occasionally
- ☐ Frequently
- ☐ Very frequently
- ☐ Almost every night

18. Over the past two weeks I have found myself feeling, or rubbing with my hand, the outside of my stomach where the baby is:

- ☐ A lot of times each day
- ☐ At least once per day
- ☐ Occasionally
- ☐ Once only
- ☐ Not at all

19. If the pregnancy was lost at this time (due to miscarriage or other accidental event) without any pain or injury to myself, I expect I would feel:

☐

Very pleased

☐

Moderately pleased

☐

Neutral (ie neither sad nor pleased; or mixed feelings)

☐

Moderately sad

☐

Very sad

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH



DASS 21 NAME _____ DATE _____

BLACK DOG INSTITUTE

Please read each statement and circle a number 0, 1, 2 or 3 which indicates how much the statement applied to you over the past week. There are no right or wrong answers. Do not spend too much time on any statement.

The rating scale is as follows:

- 0 Did not apply to me at all - NEVER
- 1 Applied to me to some degree, or some of the time - SOMETIMES
- 2 Applied to me to a considerable degree, or a good part of time - OFTEN
- 3 Applied to me very much, or most of the time - ALMOST ALWAYS

FOR OFFICE USE

		N	S	O	AA	D	A	S
1	I found it hard to wind down	0	1	2	3			
2	I was aware of dryness of my mouth	0	1	2	3			
3	I couldn't seem to experience any positive feeling at all	0	1	2	3			
4	I experienced breathing difficulty (eg, excessively rapid breathing, breathlessness in the absence of physical exertion)	0	1	2	3			
5	I found it difficult to work up the initiative to do things	0	1	2	3			
6	I tended to over-react to situations	0	1	2	3			
7	I experienced trembling (eg, in the hands)	0	1	2	3			
8	I felt that I was using a lot of nervous energy	0	1	2	3			
9	I was worried about situations in which I might panic and make a fool of myself	0	1	2	3			
10	I felt that I had nothing to look forward to	0	1	2	3			
11	I found myself getting agitated	0	1	2	3			
12	I found it difficult to relax	0	1	2	3			
13	I felt down-hearted and blue	0	1	2	3			
14	I was intolerant of anything that kept me from getting on with what I was doing	0	1	2	3			
15	I felt I was close to panic	0	1	2	3			
16	I was unable to become enthusiastic about anything	0	1	2	3			
17	I felt I wasn't worth much as a person	0	1	2	3			
18	I felt that I was rather touchy	0	1	2	3			
19	I was aware of the action of my heart in the absence of physical exertion (eg, sense of heart rate increase, heart missing a beat)	0	1	2	3			
20	I felt scared without any good reason	0	1	2	3			
21	I felt that life was meaningless	0	1	2	3			
TOTALS								

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

Five Facet Mindfulness Questionnaire

Description:

This instrument is based on a factor analytic study of five independently developed mindfulness questionnaires. The analysis yielded five factors that appear to represent elements of mindfulness as it is currently conceptualized. The five facets are observing, describing, acting with awareness, non-judging of inner experience, and non-reactivity to inner experience. More information is available in:

Please rate each of the following statements using the scale provided. Write the number in the blank that best describes your own opinion of what is generally true for you.

1	2	3	4	5
never or very rarely true	rarely true	sometimes true	often true	very often or always true

- _____ 1. When I'm walking, I deliberately notice the sensations of my body moving.
- _____ 2. I'm good at finding words to describe my feelings.
- _____ 3. I criticize myself for having irrational or inappropriate emotions.
- _____ 4. I perceive my feelings and emotions without having to react to them.
- _____ 5. When I do things, my mind wanders off and I'm easily distracted.
- _____ 6. When I take a shower or bath, I stay alert to the sensations of water on my body.
- _____ 7. I can easily put my beliefs, opinions, and expectations into words.
- _____ 8. I don't pay attention to what I'm doing because I'm daydreaming, worrying, or otherwise distracted.
- _____ 9. I watch my feelings without getting lost in them.
- _____ 10. I tell myself I shouldn't be feeling the way I'm feeling.
- _____ 11. I notice how foods and drinks affect my thoughts, bodily sensations, and emotions.
- _____ 12. It's hard for me to find the words to describe what I'm thinking.
- _____ 13. I am easily distracted.
- _____ 14. I believe some of my thoughts are abnormal or bad and I shouldn't think that way.

PRENATAL DIAGNOSIS AND MATERNAL MENTAL HEALTH

- _____ 15. I pay attention to sensations, such as the wind in my hair or sun on my face.
- _____ 16. I have trouble thinking of the right words to express how I feel about things
- _____ 17. I make judgments about whether my thoughts are good or bad.
- _____ 18. I find it difficult to stay focused on what's happening in the present.
- _____ 19. When I have distressing thoughts or images, I "step back" and am aware of the thought or image without getting taken over by it.
- _____ 20. I pay attention to sounds, such as clocks ticking, birds chirping, or cars passing.
- _____ 21. In difficult situations, I can pause without immediately reacting.
- _____ 22. When I have a sensation in my body, it's difficult for me to describe it because I can't find the right words.
- _____ 23. It seems I am "running on automatic" without much awareness of what I'm doing.
- _____ 24. When I have distressing thoughts or images, I feel calm soon after.
- _____ 25. I tell myself that I shouldn't be thinking the way I'm thinking.
- _____ 26. I notice the smells and aromas of things.
- _____ 27. Even when I'm feeling terribly upset, I can find a way to put it into words.
- _____ 28. I rush through activities without being really attentive to them.
- _____ 29. When I have distressing thoughts or images I am able just to notice them without reacting.
- _____ 30. I think some of my emotions are bad or inappropriate and I shouldn't feel them.
- _____ 31. I notice visual elements in art or nature, such as colors, shapes, textures, or patterns of light and shadow.
- _____ 32. My natural tendency is to put my experiences into words.
- _____ 33. When I have distressing thoughts or images, I just notice them and let them go.
- _____ 34. I do jobs or tasks automatically without being aware of what I'm doing.
- _____ 35. When I have distressing thoughts or images, I judge myself as good or bad, depending what the thought/image is about.
- _____ 36. I pay attention to how my emotions affect my thoughts and behavior.
- _____ 37. I can usually describe how I feel at the moment in considerable detail.
- _____ 38. I find myself doing things without paying attention.
- _____ 39. I disapprove of myself when I have irrational ideas.

Please read each statement carefully before answering. To the left of each item, indicate how often you behave in the stated manner, using the following scale:

Almost
always
5

- _____ 1. I'm disapproving and judgmental about my own flaws and inadequacies.
- _____ 2. When I'm feeling down I tend to obsess and fixate on everything that's wrong.
- _____ 3. When things are going badly for me, I see the difficulties as part of life that everyone goes through.
- _____ 4. When I think about my inadequacies, it tends to make me feel more separate and cut off from the rest of the world.
- _____ 5. I try to be loving towards myself when I'm feeling emotional pain.
- _____ 6. When I fail at something important to me I become consumed by feelings of inadequacy.
- _____ 7. When I'm down and out, I remind myself that there are lots of other people in the world feeling like I am.
- _____ 8. When times are really difficult, I tend to be tough on myself.
- _____ 9. When something upsets me I try to keep my emotions in balance.
- _____ 10. When I feel inadequate in some way, I try to remind myself that feelings of inadequacy are shared by most people.
- _____ 11. I'm intolerant and impatient towards those aspects of my personality I don't like.
- _____ 12. When I'm going through a very hard time, I give myself the caring and tenderness I need.
- _____ 13. When I'm feeling down, I tend to feel like most other people are probably happier than I am.
- _____ 14. When something painful happens I try to take a balanced view of the situation.
- _____ 15. I try to see my failings as part of the human condition.
- _____ 16. When I see aspects of myself that I don't like, I get down on myself.
- _____ 17. When I fail at something important to me I try to keep things in perspective.
- _____ 18. When I'm really struggling, I tend to feel like other people must be having an easier time of it.
- _____ 19. I'm kind to myself when I'm experiencing suffering.
- _____ 20. When something upsets me I get carried away with my feelings.
- _____ 21. I can be a bit cold-hearted towards myself when I'm experiencing suffering.
- _____ 22. When I'm feeling down I try to approach my feelings with curiosity and openness.
- _____ 23. I'm tolerant of my own flaws and inadequacies.
- _____ 24. When something painful happens I tend to blow the incident out of proportion.
- _____ 25. When I fail at something that's important to me, I tend to feel alone in my failure.
- _____ 26. I try to be understanding and patient towards those aspects of my personality I don't like.

Appendix H

REC Approval

**Health Research Authority****North West - Haydock Research Ethics Committee**

3rd Floor - Barlow House
4 Minshull Street
Manchester
M1 3DZ

Telephone: 0207 104 8012

22 February 2016

Dr Peter Taylor
Division of Clinical Psychology
Whelan Building, Quadrangle
Brownlow Hill
University of Liverpool
L69 3GB

Dear Dr Taylor

Study title:	Expecting a Baby with a Cleft: Antenatal Attachment, Mindfulness and Self-compassion
REC reference:	16/NW/0096
Protocol number:	UoL001191
IRAS project ID:	192385

The Research Ethics Committee reviewed the above application at the meeting held on 09 February 2016. Thank you for attending to discuss the application.

We plan to publish your research summary wording for the above study on the HRA website, together with your contact details. Publication will be no earlier than three months from the date of this favourable opinion letter. The expectation is that this information will be published for all studies that receive an ethical opinion but should you wish to provide a substitute contact point, wish to make a request to defer, or require further information, please contact the REC Manager Ms Rachel Katzenellenbogen, nrescommittee.northwest-haydock@nhs.net. Under very limited circumstances (e.g. for student research which has received an unfavourable opinion), it may be possible to grant an exemption to the publication of the study.

Favourable opinion

The members of the Committee present gave a favourable ethical opinion of the above research on the basis described in the application form, protocol and supporting documentation, subject to the conditions specified below. .

Conditions of the favourable opinion

The REC favourable opinion is subject to the following conditions being met prior to the start of the study.

Other ethical issues were raised and resolved in preliminary discussion before your attendance at the meeting.

Management permission must be obtained from each host organisation prior to the start of the study at the site concerned.

Management permission should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements. Each NHS organisation must confirm through the signing of agreements and/or other documents that it has given permission for the research to proceed (except where explicitly specified otherwise).

Guidance on applying for HRA Approval (England)/ NHS permission for research is available in the Integrated Research Application System, at www.hra.nhs.uk or at <http://www.rdforum.nhs.uk>.

Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of management permissions from host organisations.

Registration of Clinical Trials

All clinical trials (defined as the first four categories on the IRAS filter page) must be registered on a publically accessible database. This should be before the first participant is recruited but no later than 6 weeks after recruitment of the first participant.

There is no requirement to separately notify the REC but you should do so at the earliest opportunity e.g. when submitting an amendment. We will audit the registration details as part of the annual progress reporting process.

To ensure transparency in research, we strongly recommend that all research is registered but for non-clinical trials this is not currently mandatory.

If a sponsor wishes to request a deferral for study registration within the required timeframe, they should contact hra.studyregistration@nhs.net. The expectation is that all clinical trials will be registered, however, in exceptional circumstances non registration may be permissible with prior agreement from the HRA. Guidance on where to register is provided on the HRA website.

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

Ethical review of research sites

NHS Sites

The favourable opinion applies to all NHS sites taking part in the study taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

16/NW/0096**Please quote this number on all correspondence**

With the Committee's best wishes for the success of this project.

Yours sincerely



Dr Tim S Sprosen
Chair

E-mail: nrescommittee.northwest-haydock@nhs.net

Enclosures: List of names and professions of members who were present at the meeting and those who submitted written comments

"After ethical review – guidance for researchers"

Copy to: Alex Astor, University of Liverpool
Miss Lucy Cooper, Alder Hey Children's NHS Foundation Trust

Appendix I

Participant Information Sheet



Alder Hey Children's **NHS**
NHS Foundation Trust



UNIVERSITY OF
LIVERPOOL



Liverpool Women's
Dedicated to you



Leeds General
Infirmary

Central Manchester University Hospitals **NHS**
NHS Foundation Trust

Title of project: Antenatal Attachment, Mindfulness and Self-compassion

We would be very grateful if you would consider taking part in this research. To help you decide whether you wish to participate, please read the information below about why the research is being conducted and what it will entail. We will be happy to answer any questions you may have.

What is the purpose of the study?

This study aims to look at how 'self-compassionate', or kind, mums are to themselves in what might be a stressful, or worrying time, and how this might affect their feelings during/about the pregnancy. Additionally, the research is interested in exploring the tendency for mums to focus on the present moment, rather than thinking too much about the past or worrying about the future (this is sometimes referred to as 'Mindfulness'). We are interested in recruiting mums who are expecting a baby with a cleft lip and/or palate and mums who are **not** expecting a baby with a cleft, so this information leaflet explains the research for both these groups.

Why have I been asked to take part?

It is hoped that understanding more about expectant mums' experiences could help to tailor support received from services and increase awareness about what mums might be going through.

Do I have to take part?

No – participation is voluntary. If you decide not to take part this will not affect the care you are receiving.

What will participating in the study involve?

You will be asked to complete four questionnaires which include questions such as: "Over the past two weeks I have thought about, or been preoccupied with the baby inside me" and "I found myself getting upset by quite trivial things." The questionnaires should take approximately 20 minutes to complete. Before taking part in the study you will be asked to complete a consent form.

There is the option of completing the questionnaires on paper, electronically, or over the phone with the researcher. The paper questionnaires can either be given back to the researcher directly or posted to them at the University (stamped addressed envelopes will be provided). A webpage link will be provided if you would prefer to complete the electronic version of the questionnaires. If you would prefer to complete the questionnaires over the phone, please email, text or telephone Lisa Halpin on the contact details provided below.

Will I be reimbursed for my time?

In recognition of your time and effort in taking part you can be entered into a prize draw to win one of three £50 'LOVE2SHOP' high street shopping vouchers.

Can I withdraw from the study?

Yes – you can withdraw from the study at any time without giving a reason. You can also request for the data that you provide to be destroyed. However, if this request is received 48 hours after completing the study, this will not be possible, as your data will already have been anonymised. If you do decide to withdraw, this will not affect the care you are receiving.

What are the benefits of taking part?

The data collected from this study will hopefully give researchers and clinicians a much better understanding of expectant mums' experiences. Although the study may not directly help you, the findings will be used to support future expectant mums.

What are the possible risks of taking part?

Some people do find it difficult or upsetting to answer questions about their experiences. If you do feel affected, please discuss this with a member of the research team and we will be able to direct you to the most appropriate form of support. You will be provided with a debrief sheet which gives details of organisations that can offer support.

Will my data be confidential?

Yes – your data will be anonymised. This means that all information about you, such as your name and email address, will be removed so that you won't be identifiable. No confidential information will be included in any reporting of the study. In addition, no information will be passed on to any other person without your permission.

If you provide an email address (to receive a summary of findings or to be entered into the prize draw) this will be kept separate from your questionnaire data with no means of linking your questionnaire responses to your e-mail address. E-mail addresses will be stored securely on the University of Liverpool 'M' drive and will not be shared with any other person.

All data collected from the study will be kept safely and securely on a password protected computer. Dr Julie Van Vuuren (Lecturer in Clinical Psychology; supervising this study) will be the custodian of the study data. With your permission, the data will be archived and stored at the University of Liverpool for up to 10 years after the end of this study.

What happens when the study finishes?

After you have completed the questionnaires, you will not be asked to take part in any further research in relation to this study.

The findings of the study will be written up as part of the primary researcher's (Lisa Halpin) thesis. This forms part of her doctoral training as a Clinical Psychologist. The research will be submitted to academic journals, presented at research conferences and shared with relevant services to inform service provision.

If you are interested in the findings of the research, we will be able to send you a summary of the study once it is completed. Please select the appropriate option on the consent form if you would be interested in receiving this summary.

Who is organising and funding the study?

The University of Liverpool is the study's sponsor and has provided funding to conduct the research.

Who has given the study ethical approval?

This study has been reviewed and was given ethical approval by the Doctorate in Clinical Psychology Research Review Committee, the NHS Ethics Committee, and Research and Development approval at Alder Hey Children's Hospital, Royal Manchester Children's Hospital, Leeds General Infirmary, and Liverpool Women's Hospital.

What if I have a complaint?

If you are unhappy about any aspect of this study, please speak with the researchers. If you feel unable to do so, contact the Research Governance Officer at the University of Liverpool at ethics@liv.ac.uk or on 0151 794 8290. When contacting the Research Governance Officer, please provide details of the name or description of the study (so that it can be identified), the researchers involved, and the details of the complaint you wish to make.

Who can I contact for further information about this study?

If you have any questions, please contact:

Miss Lisa Halpin (Primary Researcher)	Lisa.Halpin.@liv.ac.uk
Dr Julie Van Vuuren (Research Supervisor)	ju1@liverpool.ac.uk
Dr Zoe Edwards (Clinical Supervisor)	0151 252 5586
Dr Rachel Mumford (Clinical Supervisor)	0151 252 5586

Thank you very much for taking the time to read this information sheet.

Appendix J
Consent Form



CONSENT FORM

Title of Research Project: Antenatal Attachment, Mindfulness and Self-compassion

Researcher: Lisa Halpin (University of Liverpool)

**Please initial
each box**

1. I confirm that I have read and understand the information sheet for the above study. I have had the chance to think about the information, ask questions and have my questions answered satisfactorily.
2. I understand that taking part is voluntary and that I can change my mind at any time without giving a reason, without my medical care or legal rights being affected.
3. I understand that my data will be anonymised 48 hours after completing the questionnaires, after which time I will no longer be able to withdraw my data.
4. I agree to my anonymised questionnaire data being stored at the University of Liverpool, in line with their policy for the storage of research data.
5. I understand that if I have provided an e-mail address, this will be stored securely (on the University of Liverpool 'M' drive) and will not be shared with another person. This will be kept separate from my questionnaire data with no means of linking my questionnaire responses to my e-mail address.
6. I understand that by initialling each point, I agree to take part in this study.

Participant Name.....

Participant Signature..... Date Signed.....

I would like to be entered into a prize draw to win a £50 high street shopping voucher YES/NO (please delete as appropriate)

I would like to receive a summary of the findings at the end of study YES/NO (please delete as appropriate)

If you have chosen YES to any of the above, please provide your email address.....